THE UNIVERSITY *of* EDINBURGH

MENTAL HEALTH, SELF-ESTEEM AND QUALITY OF LIFE OF ADULTS WITH CYSTIC FIBROSIS AND THEIR USE OF AN ONLINE DISCUSSION FORUM

Melanie Jane Platten

August 2011

Doctorate in Clinical Psychology

The University of Edinburgh

D. CLIN. PSYCHOL.
UNIVERSITY OF EDINBURGH / NHS (SCOTLAND)
TRAINING PROGRAMME

Submitted in part fulfillment of the degree of
Doctorate in Clinical Psychology at The University of Edinburgh

DECLARATION OF OWN WORK

I confirm that I have read and understood The University of Edinburgh guidelines on plagiarism and declare that this written dissertation is all my own work except where I indicate otherwise by proper use of quotes and references.

Signed: ……..

Date:……

ACKNOWLEDGEMENTS

I would firstly like to thank my supervisors Gillian Affleck, Ethel Quayle, Emily Newman and Cyan Harte who have all offered their support at different points along my thesis journey. Gillian was vital throughout the initial stages of forming my idea and navigating through ethics. Both Ethel and Emily were fantastic in providing me with guidance and constructive comments throughout data collection, analysis and write-up. Thanks also to Cyan who offered support and containment at the more fraught times. I am extremely grateful to them all for sharing their expertise and making this such an enjoyable experience.

Special thanks must be extended to the Cystic Fibrosis Trust and all the participants who chose to take part in my research. I have been moved by the resilience and strength of this group of people who face a daily battle to stay healthy. I hope that medical advances continue to be made and ever improve the lives of those affected by cystic fibrosis.

Finally, I would also like to take this opportunity to thank my family, especially my parents. I am convinced that without their support, belief and encouragement I would never have achieved what I have today. The values and work-ethic that they have instilled in me throughout my life have stood me on firm ground to enjoy all that my education and career so far has had to offer.

To everyone who has made this such a pleasure: thank you.

CONTENTS

List of tables and figures **1**
THESIS OVERVIEW **2**
ABSTRACT **3**
1 SYSTEMATIC REVIEW **5**
1.1 ABSTRACT 5
1.2 INTRODUCTION 6
1.2.1 Overview of cystic fibrosis 6
1.2.2 Health-related quality of life (HRQoL) and mental health 7
1.2.3 Social Support 8
1.2.4 Aim of review 9
1.3 METHOD 9
1.3.1 Literature search strategy 9
1.3.2 Data extraction and quality assessment 11
1.4 RESULTS 13
1.4.1 Quantitative studies 13
1.4.2 Qualitative Studies 20
1.5 DISCUSSION 21
1.5.1 Choice of measures 22
1.5.2 Quality of reported data 23
1.5.3 Review limitations 25
1.6 CONCLUSIONS 25
1.7 REFERENCES 28
2 THESIS AIMS **34**
2.1 STUDY 1 - RESEARCH QUESTIONS 36
2.2 STUDY 2 - RESEARCH QUESTION 36
2.3 ETHICAL CONSIDERATIONS 36
3 STUDY 1 METHODS **38**
3.1 DESIGN 38
3.2 PARTICIPANTS 38
3.3 PROCEDURE 39
3.4 MEASURES 41
3.4.1 Cystic Fibrosis Questionnaire – Revised (CFQ-R) 41

3.4.2 Clinical Outcomes in Routine Evaluation – Outcome Measure 34 42
3.4.3 Rosenberg self-esteem scale (Rosenberg, 1965) 42
3.4.4 Demographic questionnaire 42
3.5 DATA ANALYSIS 43
3.5.1 Quantitative data 43
3.5.2 Online resource questions 45
4 STUDY 1 RESULTS 46
4.1 DESCRIPTIVE STATISTICS 46
4.2 CORRELATIONS 47
4.3 GENDER DIFFERENCES 49
4.4 USE OF ONLINE RESOURCES 49
4.4.1 Open-ended questions 50
4.5 SUMMARY 52
5 STUDY 2 METHODS 54
5.1 DESIGN 54
5.2 PROCEDURE 54
5.3 PARTICIPANTS 54
5.4 DISCUSSION FORUM 55
5.5 DATA ANALYSIS 55
5.5.1 Qualitative data 55
6 STUDY 2 RESULTS 60
6.1 REGULAR OR DAILY CF TREATMENT 60
6.2 HOSPITAL EXPERIENCES AND PROCEDURES 64
6.2.1 Hospital experiences 64
6.2.2 Medical procedures 65
6.3 CHANGES IN HEALTH AND PROGRESSION OF ILLNESS 67
6.3.1 Changes in health 67
6.3.2 Physical development and growth 68
6.3.3 Lung transplant 69
6.4 DAILY LIVING AND OCCUPATION 71
6.4.1 Education and employment 71
6.4.2 Disability Living Allowance (DLA) and benefits 72
6.4.3 Travel and healthcare abroad 73
6.4.4 Other topics 73

6.5 PSYCHOSOCIAL PROCESSES AND ISSUES 74
6.5.1 Adjustment and transitions 74
6.5.2 Interpersonal relationships 75
6.5.3 Emotions, coping and psychology services 76
6.5.4 Body image and weight 77
6.5.5 Social support 78
6.6 FUTURE 79
6.6.1 Fear and hope 79
6.6.2 Fertility, pregnancy and parenting with CF 80
6.6.3 Future changes in the NHS and DLA 81
6.7 TONE OF POSTS AND PARTICIPANT NARRATIVES 81
6.8 SUMMARY 82
7 DISCUSSION 84
7.1 MENTAL HEALTH, QUALITY OF LIFE AND SELF-ESTEEM 84
7.2 CORRELATIONS BETWEEN VARIABLES 87
7.3 USE OF ONLINE FORUMS 89
7.4 LIMITATIONS 94
7.5 ETHICAL CONSIDERATION OF ONLINE RESEARCH 96
7.6 CLINICAL IMPLICATIONS 98
7.7 DIRECTIONS FOR FUTURE RESEARCH 100
8 JOURNAL ARTICLE 103
ABSTRACT 104
Key words: 104
INTRODUCTION 104
METHODS 106
DESIGN 106
PARTICIPANTS 106
PROCEDURE 107
MEASURES 109
Cystic Fibrosis Questionnaire – Revised (CFQ-R) 109
Clinical Outcomes in Routine Evaluation – Outcome Measure 34 (CORE-OM) 109
Rosenberg self-esteem scale (24) 109
Demographic questionnaire 110
DATA ANALYSIS 110

Correlations 110
ETHICAL APPROVAL 111
RESULTS 111
DESCRIPTIVE STATISTICS 111
CORRELATIONS 112
GENDER DIFFERENCES 112
DISCUSSION 114
Limitations 115
Clinical implications and future research 116
REFERENCES 118
9 REFERENCES 122
10 APPENDICES 133
APPENDIX A: SYSTEMATIC REVIEW PROFORMAS 134
APPENDIX B: ETHICS APPROVAL LETTER 140
APPENDIX C: CF TRUST AGREEMENT EMAILS 142
APPENDIX D: CLINICAL PSYCHOLOGY REVIEW AUTHOR GUIDELINES 144
APPENDIX E: COPY OF ONLINE SURVEY FOR STUDY 1 148
APPENDIX F: EMAIL PERMISSIONS FOR STANDARDISED MEASURES 161
APPENDIX G: INITIAL INDEX FRAMEWORK FOR QUALITATIVE ANALYSIS 165
APPENDIX H: FINAL INDEX FRAMEWORK FOR QUALITATIVE ANALYSIS 167
APPENDIX I: EXAMPLE OF A FRAMEWORK ANALYSIS DATA MATRIX 168
APPENDIX J: JOURNAL OF CYSTIC FIBROSIS AUTHOR GUIDELINES 169

List of tables and figures

Table 1.1 Inclusion and exclusion criteria for systematic review articles 11

Table 1.2 Summary of quantitative papers included for review 14

Table 3.1 Number of survey sections completed out of 105 consenting participants..... 38

Table 3.2 Key demographic characteristics of full and partial surveys completed 39

Table 3.3 Demographic and basic clinical information summary for full data sets 40

Table 4.1 Means and standard deviations of outcome measure scores by gender 46

Table 4.2 Correlation coefficients and significance for outcome measures 48

Table 4.3 Uses of discussion forum (N = 64) and Facebook page (N = 40) 50

Table 5.1 Example process for arriving at a theme 58

Figure 1.1 Flowchart of systematic review paper selection process 12

Figure 6.1 Main themes resulting from framework analysis of discussion forum posts.. 61

Word count: 27, 867

THESIS OVERVIEW

This thesis follows the portfolio model of work carried out in part-fulfilment of the Doctorate in Clinical Psychology at the University of Edinburgh. An abstract provides an overview of the whole thesis, its findings and implications. Chapter 1 consists of a systematic review looking at the published research into the mental health and social support in adolescents and adults with cystic fibrosis (CF). This review is written for submission to Clinical Psychology Review for publication and follows their author guidelines. Chapter 2 links the systematic review to the thesis project and outlines its aims and research questions. The main project is then presented as two separate studies. Study 1 is a quantitative assessment of the mental health, quality of life and self-esteem in adults with CF and the relationships between these factors. The methods and results for this study are presented in chapters 3 and 4 respectively. Chapters 5 and 6 present the methods and results of Study 2. This is a qualitative exploration of the way adults with CF make use of an online discussion forum. The findings of studies 1 and 2 are brought together within chapter 7 which forms the thesis discussion. This chapter contains consideration of the results in the context of existing published literature, their limitations and clinical implications. Chapter 8 presents Study 1 written up in the form of a journal article. This follows author guidelines for submission to the Journal of Cystic Fibrosis. The final two sections of the thesis portfolio include the references (chapter 9) and appendices (chapter 10).

ABSTRACT

Background: Improvements in cystic fibrosis (CF) treatment over the last two decades have brought large increases in life expectancy. As a result, researchers have become more interested in the long-term quality of life and psychological wellbeing of adults with CF. Introduction of segregation policies to prevent cross-infection have added a new dimension to life with CF; patients are discouraged from meeting face-to-face and this impacts on access to peer support.

Systematic review: A systematic search of the published literature since 1990 identified 10 journal articles describing research with adolescents and adults with CF, their mental health needs and social support or functioning. Quantitative papers indicated an inverse relationship between mental health and social support or social functioning.

Objectives: The internet has become increasingly used for social networking and accessing social support resources. This thesis aimed to explore the psychological wellbeing of adults with CF who make use of online social support and the themes emerging within online discussion forums.

Method: Study 1 is a quantitative survey carried out online which assesses the quality of life, mental health and self-esteem of 74 adults with CF. Study 2 uses framework analysis to provide a qualitative exploration of posts made by participants on an online discussion forum.

Results: Moderate levels of quality of life and comparably high levels of self-esteem were found. Thirty per cent of the sample population scored above threshold for clinical levels of mental health difficulty. Significant correlations indicated that poorer self-esteem was associated with poorer quality of life and greater mental health difficulty. Six themes emerged from the

discussion forum posts including CF treatment, daily living and occupation, health exacerbations, psychosocial processes and issues and the future.

Conclusions: Self-esteem and mental health scores were similar to previous research. However, quality of life was noted to be considerably lower when compared to other research samples. This was thought to be influenced by the overrepresentation of females in the current sample but may also be reflective of the group of people with CF who make use of online resources. Results are discussed in relation to clinical implications, limitations and directions for future research, including the efficacy of internet groups in improving wellbeing and consideration of possible developments in technology to promote socialisation in the CF population.

1 SYSTEMATIC REVIEW

1.1 ABSTRACT

Cystic fibrosis (CF) has considerable impacts on socialisation due to treatment demands and segregation policies designed to protect against physical health decline. The prevalence of mental health difficulties within the CF population has been shown to vary with age, gender and work status. They also impact negatively on quality of life and treatment adherence. Few studies include consideration of how social support influences psychological well-being in people with CF. The current review aims to examine the findings of published literature which looks at the relationship between mental health and social support in adolescents and adults with CF. Online database and hand searches of relevant journals led to the identification of 9 articles eligible for review. Quantitative papers supported an inverse relationship between mental health symptoms and social support. Factors which may influence this relationship are discussed, including severity of illness. The findings are discussed with consideration for statistical weaknesses and future research possibilities.

Key Words: cystic fibrosis, mental health, social support, systematic review

Highlights:

- Systematic review of papers considering mental health and social support in people with CF
- Few papers exist which examine this relationship
- Available research suggests a negative correlation between the two variables
- Online social environments may help people with CF overcome segregation policies to enhance peer support

1.2 INTRODUCTION

1.2.1 Overview of cystic fibrosis

Over 8500 people in the UK suffer from cystic fibrosis (CF), one of the most common life-threatening inherited diseases in the UK (CF Trust, 2011). CF primarily affects the lungs and digestive system (Ernst, Johnson, & Stark, 2010) whilst additional problems with liver function, fertility, bone density or CF-related diabetes (CFRD) may develop throughout the life-course (Ernst, et al., 2010). Advances in treatment have led to drastic increases in median life expectancy, which is currently reported to be 38 years old (CF Trust, 2011). However, daily treatment regimes for CF are time-consuming and complex, often taking between 2 to 4 hours each day (Quittner, Barker, Marciel, & Grimley, 2009). Ordinarily, treatment consists of a combination of medication (e.g. inhaled antibiotics, oral digestive enzymes), high calorie diet, chest physiotherapy and exercise. These treatments help to mobilise thick secretions of mucus in the airways, improve the gas exchange processes between the lungs and blood stream and improve the absorption of nutrients in the digestive system. In addition, patients often receive intravenous antibiotics as hospital inpatients to treat respiratory infections.

The build up of mucus in the lungs makes people with CF more vulnerable to infections which can have detrimental consequences for lung function and physical health. The frequency of infections and exacerbations often increases with age and results in greater treatment burden and stress (Ernst, et al., 2010). In an attempt to minimise the cross-infection of particularly harmful bacteria, such as *pseudomonas aeruginosa*, segregation policies were introduced in the early 1990's to separate CF patients on the grounds of their infection status (Littlewood, 2004). Strains of these infections acquired from fellow patients are harder to treat than those contracted from the environment (CF Trust, 2011). Although the physical benefits are argued to outweigh

the psychological impact (Geddes, 2001), segregation policies have reduced the opportunity for people with CF to meet and interact with their CF-peers whilst increasing stigma and isolation experienced by patients (Quittner, et al., 2009; Russo, Donnelly, & Reid, 2006).

1.2.2 Health-related quality of life (HRQoL) and mental health

With the vast majority of people with CF now living into adulthood, researchers have become increasingly interested in long-term quality of life and psychological well-being. A past review paper by Pfeffer and colleagues (Pfeffer, Pfeffer & Hodson, 2003) provided an overview of research pertaining to psychological aspects of CF, including mental health and quality of life. Their paper recognised the variability in findings regarding factors which may be influential for the mental health of people with CF, such as gender, age, coping style and severity of illness. Although the review is informative and appears to provide a summary of some key factors involved in the psychosocial aspects of cystic fibrosis, it appears not to have been conducted in a systematic manner. The authors did not report their strategy for searching the literature nor whether the quality of papers was considered in the synthesis of their findings.

Since Pfeffer, et al.'s (2003) review, there have been numerous publications which further explore the quality of life and mental health of people with CF. Studies have indicated that adolescents and adults with CF report high levels of quality of life despite the large treatment demands and shortened life-span in CF (Ernst, et al., 2010; Szyndler, Towns, van Asperen, & McKay, 2005). On the contrary, research studies into rates of mental health symptoms have presented a more variable picture. Some studies suggest higher levels of depression and/or anxiety compared to the general population (Bregnballe, Thastum, & Schiøtz, 2007; Goldbeck et al., 2010; Modi, Driscoll, Montag-Leifling, & Acton, 2011; Quittner et al., 2008), whereas others maintain that levels of mental health symptoms are comparable to the general population

(Anderson, Flume, & Hardy, 2001; Szyndler, et al., 2005). The reasons for this variability continue to be explored by research with recent influential factors shown to include gender, age, work status and severity of illness (Bregnballe, et al., 2007; Burker et al., 2004; Goldbeck, et al., 2010; Modi, et al., 2011; Pearson, Pumariega, & Seilheimer, 1991; Quittner, et al., 2008).

1.2.3 Social Support

The research literature has frequently acknowledged the time-consuming nature of CF treatment regimes and, intuitively, that time spent on treatment decreases the time available to engage in other activities such as socialising (Modi, et al., 2011). Peer relationships have been seen to be affected in other chronic illnesses (Forgeron et al., 2010) and, with the use of segregation in CF services, it is important to consider how people with CF can build and maintain relationships with their CF peers (Quittner, et al., 2009; Russo, et al., 2006). Quality of life measures usually include assessment of social *functioning* (Cella, 1998; Spilker, 1996) but this may be conceptually different to social *support*. The term *social functioning* suggests looking at how well people are able to take part in social activities or interactions. *Social support*, however, suggests looking at the availability of support which is sought from or provided by family, friends, health professionals or work colleagues. In the general population without chronic health conditions, higher levels of social support are important for adaptation to stress and are related to lower levels of mental health difficulty (Cohen & Wills, 1985; Coyne & Downey, 1991; Hefner & Eisenberg, 2009; Holahan & Moos, 1981). In papers specifically focusing on social support in CF, peer relationships have been shown to be important in buffering against the negative effects of poor family relationships (Herzer, Umfress, Aljadeff, Ghai, & Zakowski, 2009). In addition, social support from others, including family members and friends, is related to better coping and improved adherence to CF treatment (Barker, 2010; White et al., 2009). Key researchers have suggested that future studies examine the potential buffering effects that social

support may have on depression in CF patients (Modi, et al., 2011) and explore how new technologies may enhance peer interaction for this group (Quittner, et al., 2009).

1.2.4 Aim of review

In light of the potential importance of social support for patients with CF and its likely influence in mental health and psychological well-being, it was considered that a review of the published literature in this area may be beneficial. Pfeffer, et al.'s (2003) review briefly mentioned the possible impacts of social support on mental health but this was not a key focus of their paper. They provided a much wider summary of research into the psychosocial aspects of CF published since 1990 (Pfeffer, et al., 2003), but it was unclear whether papers were identified or approached in a systematic manner. In order to provide a more focussed and systematic examination of the literature which considers relationships between social support and mental health in people with CF it was felt that a further review would be beneficial. Given the paucity of research using specific measures of social support, papers including measures of social functioning were also of interest. Therefore, the aim of this current review was to provide a systematic evaluation of research published since 1990 which considers the importance of and possible relationships between social support and mental health in adolescents and adults with cystic fibrosis.

1.3 METHOD

1.3.1 Literature search strategy

EMBASE, PsycINFO and MEDLINE online databases were searched between January 1990 and January 2011. These databases were chosen to cover a range of online journals from the social sciences, nursing and medical disciplines. Titles, abstracts and keywords were searched using the following terms (cystic fibrosis) AND (social support OR peer support OR peer relationships

OR friendships) AND (mental illness OR mental health OR mental disorder OR psychosocial OR self-esteem OR anxiety OR depress* OR psychological wellbeing). A manual search of the contents pages of four keys journals, namely the Journal of Pediatric Psychology, Journal of Cystic Fibrosis, Chest and Journal of Health Psychology, was also conducted over the same time period to ensure the inclusion of key papers which may have been missed by electronic database searches.

Search results were assessed using predetermined inclusion and exclusion criteria (see Table 1.1). In the first instance, titles and citations of papers were screened for the presence of exclusion criteria. Those meeting at least one of these exclusion criteria were discarded. For the remaining papers, abstracts were assessed for inclusion. In order to ascertain a level of criteria reliability, an independent clinical psychologist experienced in qualitative and quantitative research was involved in the assessment process. Due to time limitations, it was not possible for all abstracts to be assessed by a second researcher, therefore a random sub-set of 20 abstracts were rated using the protocol and criteria developed *a priori*. Agreement on eligibility was found for 19 out of 20 papers. Three of the 19 papers were obtained in full-text to clarify eligibility because both reviewers felt that a decision could not be made about inclusions based on the abstract alone. The single discrepancy applied to a reference for a conference abstract; initial criteria were revised to ensure this was identified as a reason for exclusion.

Full-text articles were obtained for studies which could not be excluded on the basis of the abstract. Final assessment of eligibility was carried out on full-text articles and those meeting criteria were included in the review. In order to identify additional papers which may have been missed from electronic searches two strategies were used: reference lists of identified journal articles were hand-searched and papers citing non-excluded studies were checked for eligibility.

In total, 23 full-text papers were obtained for final assessment for inclusion by the primary researcher. Of these papers, 14 did not meet inclusion criteria; reasons included lacking a mental health measure, no consideration of social support or focusing on parents and siblings without CF. The selection process is illustrated in Figure 1.1 and provides the number of studies discarded at each phase and reasons for rejection.

Table 1.1 Inclusion and exclusion criteria for systematic review articles

Inclusion	**Exclusion**
▪ Participants have CF ▪ 13 years old + ▪ Peer-reviewed research articles ▪ English language ▪ Measurement or report of mental health ▪ Measurement or report of social support or social functioning	▪ Participants are: • Children with CF <13 years old • Parents or carers without CF • Adolescents or adults without CF ▪ Non-peer reviewed research: • Book chapters • Book reviews • Conference Abstracts • Letters • Theses/Dissertations ▪ Non-English language article

1.3.2 Data extraction and quality assessment

A standard proforma for assessment of article quality and data extraction was developed in consultation with a research colleague. Both quantitative and qualitative papers were assessed

for methodological rigour and quality. Checklists provided by the Scottish Intercollegiate Guidelines Network within 'SIGN 50: A guideline developer's handbook' (2008) were adapted to provide relevant critique of the design and quality of quantitative papers as eligible. The SIGN 50 quality criteria checklists are based on the MERGE (Method for Evaluating Research and Guideline Evidence) checklists developed in Australia (Liddle, Williamson, & Irwig, 1996). These guidelines have been used internationally for systematic reviews published in peer-reviewed journals (e.g. Glare, et al., 2004; Ishihara & Brayne, 2005; Liang, et al., 2010; Soon, et al., 2009). Qualitative papers were scrutinised using an alternative framework from the Critical Appraisal Skills Programme [CASP] (Public Health Resources Unit, 2006) with additional data extraction questions. A copy of both frameworks can be found in Appendix A.

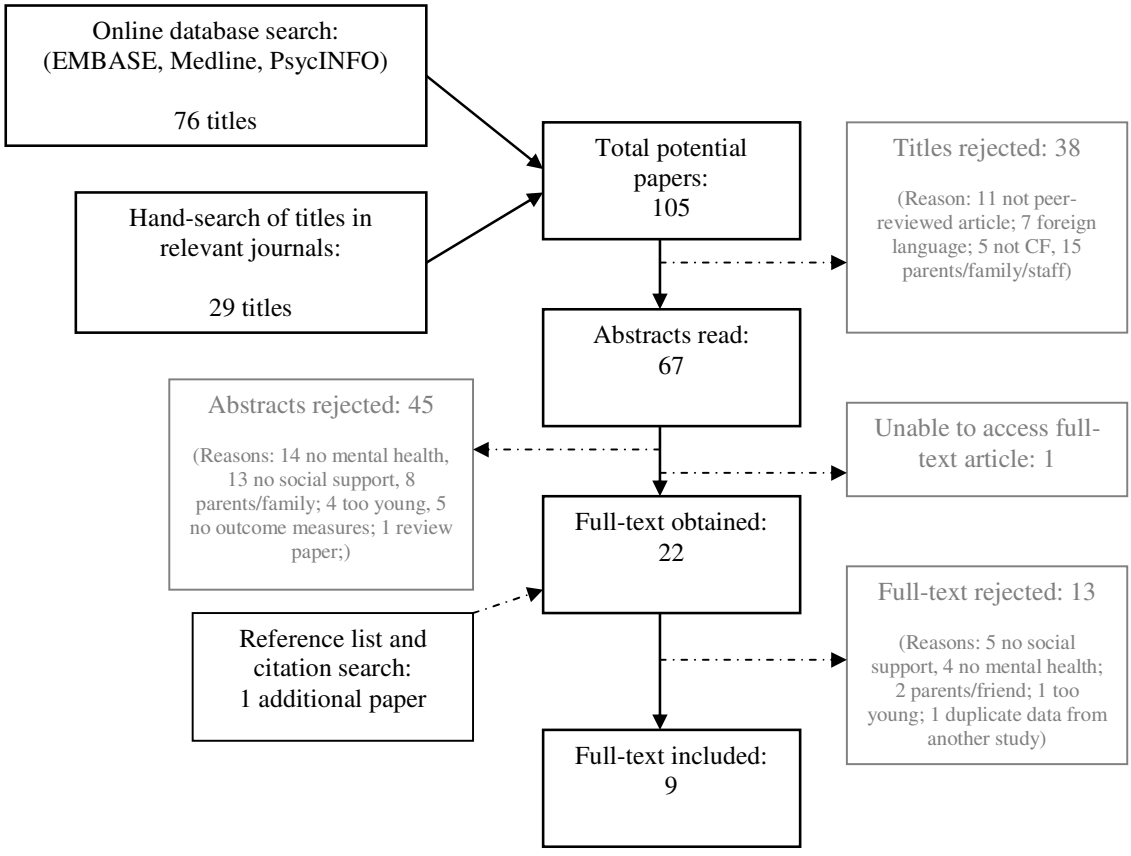

Figure 1.1 Flowchart of systematic review paper selection process

1.4 RESULTS

Of the 9 papers identified and included in the final review, 7 were quantitative and 2 were qualitative in nature. These papers will be summarised using a narrative approach, considering quantitative papers in the first instance, followed by the qualitative papers.

1.4.1 Quantitative studies

The seven quantitative papers deemed eligible for inclusion in the review varied in focus and the majority took a cross-sectional approach to assessing variables within a population of patients with CF. One exception to this was Casier's (2011) prospective study of the role of acceptance in well-being. An overview of the studies is provided in Table 1.2. Data outlining each study's sample is given where possible. Where available, this includes report of FEV_1% predicted (forced expiratory volume in 1 second, per cent predicted) which is a frequently used measure of lung function and indicator of illness severity where higher percentages indicate better respiratory functioning. The papers will be summarised in terms of their mental health and social support findings and then the relationships found between these two factors. It is acknowledged that eligible papers do not represent the wider research literature in the area of mental health needs in the CF population, but it was considered important to summarise the findings within each of these areas before joint consideration.

1.4.1.1 Mental health and psychological well-being

The papers within the selected sample provide some insight into the mental health of people with CF using a variety of assessment measures. Anderson et al. (2001) aimed to report a baseline

Table 1.2 Summary of quantitative papers included for review

Authors	Brief Description	Sample	Measures	Statistics	Main Conclusions
Anderson et al. (2001)	Assesses psychological profiles of adults with CF and predictors of good mental health	N = 34 (mean age = 28.5, SD = 8.0); 59% male; mean FEV_1 = 47.9%	BDI; MMPI-2; STAI; MHLOC; Perceived Social Support	T-tests MANOVA Regression analyses	Psychopathology at comparable level to general population. No significant differences in scores compared on gender, age or FEV_1. MANOVA showed better psychological functioning was predicted by being female, having higher levels of social support and better health status.
Burker et al. (2000)	Assesses QoL in people awaiting lung transplant. Compared those with CF with other end-stage lung disease	CF: N = 58 (mean age = 27.42, SD = 6.23), 49% male Other disease: N = 52 (mean age = 45.52, SD = 9.55)	BDI; STAI; SIP; MOS-SSS; COPE Inventory	ANOVAs (with age as covariate)	Compared to non-CF group, the CF group showed significantly: lower anxiety, similar levels of depression, had greater availability of social support (although not when age taken into account), more likely to accept their lung-disease. Significant negative correlation between anxiety and social support in CF group.
Casier (2011)	Prospective study of the role of acceptance in the well-being of adolescents and adults with CF	Time 1: N = 40, 58% male. Time 2: (6 months later) N = 28 (mean age = 18.7 years, SD = 2.8) mean FEV_1 = 83.0%	ICQ; CFQ; HADS	Pearson's r correlations T-tests Hierarchical regressions	Time 1: Greater acceptance related to less anxiety, less depression, better social, emotional, role functioning. Time 2: Greater acceptance related to less depression. Anxiety and depression were negatively correlated with social functioning at T1 and T2.
Delelis et al. (2008)	Explores marital satisfaction, emotional experiences and coping in adults with CF	N =16 (+ 16 partners) (mean age = 28 years, SD = 4.56) 50% male 9 ‘minor’ and 7 ‘major’ severity of CF	STAI; CES-D; DAS-16; WCC	ANOVAs Pearson's r correlations Mann-Whitney	Patients with higher depression or anxiety showed lower marital satisfaction and more emotion-focused coping strategies. Patients classified as major severity used more emotion-, problem- and social support-focused coping than those with minor severity. 31% and 25% patients score above cut-off for anxiety and depression respectively.

Authors	Brief Description	Sample	Measures	Statistics	Main Conclusions
Havermans et al. (2008)	Presents assessment of the association between QoL and anxiety or depression in adults with CF	N = 57 (mean age = 26.79, SD = 8.15) 51% male mean FEV_1 = 65.1%	CFQ (Dutch); HADS	ANOVAs Pearson's r correlations	Mean HADS scores comparable to general population. High anxiety scores associated with poorer scores on CFQ domains including social functioning. Depressive symptoms related to lower scores on CFQ domains excluding social functioning. Significant relationships remained when controlling for FEV_1.
Riekert et al. (2007)	Cross-sectional evaluation of relationship between lung function, QoL and depression in adults with CF	N = 76 (mean age 30.6 years, SD = 9.6) 44.7% male mean FEV_1 = 62.8%	CFQ; BDI	Spearman's Rho Kruskal-Wallis test with Bonferonni corrections	Depressive symptoms present in 30% of sample. Similar HRQoL to other studies of CF. Higher depression scores associated with lower FEV_1 and all CFQ subscales. Those with depressive symptoms had poorer HRQoL scores regardless of lung function.
Szyndler et al. (2005)	Exploration of family and psychological functioning and QoL in adolescents with CF	N = 52 (mean age 15.06 years, SD = 1.98) 51.9% male mean FEV_1 = 72.4%	CFQ; HOPES; FES; SCL-90-R	Spearman's Rho Mann-Whitney U	Levels of mental health need lower than general population. Global severity index on SCL-90-R correlated negatively with scores on social subscale of CFQ. Cohesiveness, expressiveness and organisation subscales on the FES were positively correlated with SCL-90-R global score. Also positive correlation between family cohesion and emotional functioning subscale of CFQ.

Abbreviations: BDI = Beck Depression Inventory; CES-D = Centre for Epidemiologic Studies - Depression Scale; CFQ-R = Cystic Fibrosis Questionnaire; DAS-16 = Dyadic Adjustment Scale; FES = Family Environment Scale - 3rd edition; FEV_1 = forced expiratory volume in 1 second % predicted; HADS = Hospital Anxiety and Depression Scale; HOPES = Hunter Opinions and Personal Expectations Scale; ICQ = Illness Cognition Questionnaire; MHLOC = multidimensional health locus of control; MMPI-2 = Minnesota Multiphasic Personality Inventory; QoL = quality of life; MOS-SSS = Medical Outcomes Study – Social Support Survey; SCL-90-R = Symptom Checklist-90-Revised; SIP = Sickness Impact Profile; STAI = Spielberger State-Trait Anxiety Inventory; WCC = Ways of Coping Checklist

assessment of the psychological needs of the adult CF population and explore predictive variables associated with better psychological functioning. In their sample of 34 adults with CF, they discovered levels of depressive symptoms and anxiety which were comparable to the general population. Subsequent papers in the current review reported similar findings (Casier, et al., 2011; Havermans, Colpaert, & Dupont, 2008; Szyndler, et al., 2005). A slightly earlier paper by Burker, Carels, Thompson, Rodgers and Egan (2000) found that a sample of patients with CF awaiting lung transplantation had lower levels of mental health difficulty than those with other end-stage lung diseases also awaiting transplant. In contrast, an interesting paper by Delelis, Christophe, Leroy, Vanneste and Wallaert (2008) examined the psychological functioning, coping and marital satisfaction in sixteen couples in which one person had CF. Although the sample size was small, the authors found that there were no differences in the levels of mental health symptoms between patients with CF and their healthy partners. Both patients and partners experienced higher levels of anxiety than depression. Despite the lack of difference between partners and patients, the proportion of participants (both patients and partners) experiencing clinical levels of anxiety or depression was higher than indicated in the general population. Five out of the 16 patients (31 per cent) and 7 out of 16 (44 per cent) partners scored above the cut off for significant anxiety symptomology. For depressive symptoms, 4 patients (25 per cent) and 2 partners (12.5 per cent) scored above cut-offs for possible cases of depression. Rates of anxiety and depression in the general population are suggested to be in the region of 16 per cent (Singleton, Bumpstead, O'Brien, Lee, & Meltzer, 2001).

A larger study also identified elevated levels of depression amongst a sample of 76 adults with CF (Riekert, Bartlett, Boyle, Krishnan, & Rand, 2007). Whilst exploring the relationships between quality of life and mental health, Riekert et al. (2007) noted the presence of depressive symptoms in 30 per cent of their sample using the Beck Depression Inventory [BDI] (Beck,

Steer, & Barbin, 1988). However, this finding was not replicated within a study by Havermans et al. (2008) who aimed to further explore the relationships between mental health and lung functioning using the Hospital Anxiety and Depression Scale [HADS] (Zigmund & Snaith, 1983). The exact reason for the contrast between Riekert et al.'s (2007) and Delelis et al.'s (2008) findings compared with other studies is not clear but it may be due to the assessment measures used, sample size, age of participants or factors which are not apparent from the reported methodology.

1.4.1.2 Social support and social functioning

Only two papers identified for review made use of standardised measures designed explicitly to measure social support (Anderson, et al., 2001; Burker, et al., 2000). Another two papers included consideration of social support through using measures of coping and family environment (Delelis, et al., 2008; Szyndler, et al., 2005). Delelis et al. (2008) found that, in their small sample of participants with CF, seeking social support was used as a coping strategy more often in those with a more severe classification of CF. The remaining papers mentioned social aspects of their participants' lives through the use of the social functioning subscale of the Cystic Fibrosis Questionnaire - Revised [CFQ-R] (Quittner, Buu, Watrous, & Davis, 2002). In general, papers suggested that CF patients have high levels of perceived social support (Anderson, et al., 2001; Burker, et al., 2000) and social functioning (Casier, et al., 2011; Havermans, et al., 2008; Riekert, et al., 2007; Szyndler, et al., 2005) despite potential difficulties in interpersonal relationships (Pfeffer, et al., 2003).

1.4.1.3 Relationships between mental health and social support

Although not all papers carried out correlations between measures of mental health and social support (e.g. Delelis et al., 2008), the six papers which did pointed towards a negative

correlation between mental health difficulty and social support or functioning. Higher levels of social support were related to lower levels of depression (Anderson, et al., 2001) and anxiety (Anderson, et al., 2001; Burker, et al., 2000). Similarly, higher levels of social functioning were related to lower levels of anxiety (Casier, et al., 2011; Havermans, et al., 2008), depression (Casier, et al., 2011; Riekert, et al., 2007) and global measures of mental health symptoms (Szyndler, et al., 2005). People with CF living in a supportive family environment, in particular families with higher levels of cohesion, expressiveness and organisation, appeared to have lower levels of psychopathology and better emotional functioning (Szyndler, et al., 2005). The types of coping used by adults with CF differed depending on levels of depression and anxiety: those with higher levels of anxiety or depression relied on emotional coping strategies rather than social support strategies (Delelis, et al., 2008).

Due to the nature of assessing relationships using correlations of cross-sectional data, it is not possible to infer causality. As a consequence, it is unclear whether higher levels of social support or social functioning serve as a protective factor against developing mental health difficulties or whether the development of mental health difficulties reduces the levels of social functioning or perceived social support. Longitudinal studies would be required in order to make inferences about the causality in this relationship.

1.4.1.4 Additional influential variables

The studies mentioned within this review present evidence for a negative correlation between mental health symptoms and social support and functioning. However, a number of the studies reveal that this relationship is likely to be complex and influenced by a number of other variables. Physical health status or disease severity, gender and level of acceptance were noted in relation to some findings.

It has been shown that when physical health status is factored into data analysis, significant relationships between mental health and social functioning scores persist (Havermans, et al., 2008; Riekert, et al., 2007). This indicates that physical health status does not account for the relationship between social support and mental health but it seems that social support may act as a moderator for the relationship between physical health status and mental health. In Anderson et al.'s (2001) paper, regression analyses revealed better psychological functioning to be predicted not only by higher levels of social support but also by being female and being in better physical health. Reikert et al. (2007) also noted a significant relationship between better lung functioning, fewer depressive symptoms and better quality of life. Moreover, they also showed that those with poorer lung function (FEV_1% predicted < 70%) had three times the odds of scoring above the clinical cut-off score on the BDI. A further paper highlighting the influences of physical health status was that by Delelis et al. (2008) who noted the potential impact of physical health status in the differential use of coping strategies employed depending on CF severity. Those who were rated as having "major" CF (based on a number of physical health indicators) were seen to identify social support as a coping strategy significantly more than those rated with "minor" CF. The greater identification of social support as a coping strategy in those with major severity CF may be a possible reason why levels of anxiety or depression were no higher in this group compared to the minor CF severity group

A final influential factor is level of acceptance. Casier et al.'s (2011) study indicated the important role of acceptance in the symptoms of mental health at two time points six months apart. They found that higher levels of acceptance were associated with fewer depressive symptoms at both time points. Social functioning, as well as other sub-domains of quality of life measured by the CFQ-R, was related to acceptance at the first time point. No relationship with

acceptance was found for anxiety. The authors acknowledge that their small sample size may have led to small or medium effects within data being overlooked and that further research is required. However, their study suggests that acceptance may play a protective role for patients with CF and that it perhaps has a positive influence on mental health and quality of life, including social functioning.

1.4.2 Qualitative Studies

Two qualitative studies were identified as eligible for inclusion within the present systematic review (Durst et al., 2001; Macdonald, 2006). Both considered the experience of CF patients in relation to lung transplantation, but where Macdonald (2006) focused on the period of time waiting for lung transplantation, Durst et al. (2001) explored experiences post-transplantation. Macdonald (2006) presents a clear and robust methodology involving semi-structured interviews with eight adult CF patients, 4 pre- and 4 post-lung transplant. The author's content analysis revealed four themes running through the interview data: displacement, disorder, life in limbo and readjustment to wellness. In relation to the current review, Macdonald's (2006) participants described experiencing "disordered emotions" whilst attending assessment for lung transplant and when adjusting after transplant. The need for emotional and practical support from loved-ones, carers or other post-transplant peers was highlighted as important when coping with emotional difficulties triggered by the process.

Durst et al.'s (2001) study similarly considered psychological functioning and social aspects of life for CF patients in relation to lung transplantation. Their cohort of 19 adolescents who had undergone transplant was interviewed on topics including psychological and social functioning. The paper reports that adolescents functioned well in terms of their psychological and emotional well-being. Participants reported feeling that transplant had impacted positively on their self-

esteem, a variable which is often linked with better mental health (Mann, Hosman, Schaalma, & de Vries, 2004). Interviews also revealed that participants felt their transplant had improved their social interactions with peers, which in turn may influence accessibility of social support. However, in contrast to Macdonald (2006), Durst et al.'s (2001) report of their qualitative methodology and data analysis strategies lacked detail and made no reference to the methods used. Although this article sheds light on the experiences of adolescents with CF post-transplant, the quality assessment of the paper indicated a marked lack of scientific rigour. There was no consideration of potential biases such as: the influence of interviewer, the context in which the questions were being asked, the method of analysis used or the possible influence of researcher bias during the analysis process. The poor presentation of the methods compromises the validity and robustness of the findings.

The qualitative studies fit well with the quantitative papers discussed previously. Macdonald's (2006) study indicated that patients required social support when going through the transplant process, a time when lung functioning and physical health are particularly poor, in order to cope more effectively with emotional difficulties. Durst et al. (2001) describe links between better physical health, provided by the lung transplant, increased self-esteem and social interaction. These factors perhaps all contribute to better psychological well-being.

1.5 DISCUSSION

Relatively few published studies were found which looked at mental health and its relationship with social support in adolescents and adults with CF. Identified studies which met criteria suggested an inverse relationship between social support and mental health symptoms, i.e. those with greater social support or social functioning appear to experience lower levels of psychological distress or mental health difficulty. The complexities of this relationship are yet to

be clarified and it is still not clear to what extent some factors such as disease severity and acceptance play a role. It may be that these factors moderate the relationship, or alternatively, that social support moderates the relationship between disease severity and psychological well-being.

Whilst carrying out the review, two areas for consideration came to light, namely choice of outcome measures and quality of papers, and these are discussed below.

1.5.1 Choice of measures

The studies included within the review employed a number of different measures to assess mental health and social support. It is understood that the choice of assessment measures depends on variables such as availability of measures within services, financial resources, researcher preference and the specific target variables within a project. The development of a reliable and valid CF-specific quality of life measure, namely the CFQ-R (Quittner, et al., 2002) has been beneficial to the research area. It provides insight into quality of life in several different domains including physical health, emotional functioning and social functioning. It is becoming more frequently used in projects and this enables some level of comparison across studies. However, despite inclusion of an emotional functioning sub-domain, it does not provide measurement of mental health symptoms per se and the choice of an additional measure for this purpose seems to vary a great deal. Within the seven quantitative papers included in this review, five different measures of mental health were used (BDI, HADS, CES-D, SCL-90-R, STAI), only some of which were reported to be reliable and valid within a physical health population. It is possible that some of the psychological difficulties faced by people with CF, such as adjustment to diagnosis, coping with lung transplantation and end-of-life issues, may not be accurately captured by current measures of mental health. As more research is carried out into

the psychological needs of the CF population reliability and validity of existing measures may well be established but, for now, it is important to acknowledge their possible limitations.

As a consequence of considering different measures used within the identified studies it was recognised that social aspects of life for patients with CF were reported in terms of social functioning or social support. These two terms seem to be used interchangeably within some research but may actually have important conceptual differences as outline in the introduction. Within this review, social support and functioning have been considered as two different concepts but, of course, social functioning may be a prerequisite for being able to access or benefit fully from social support. In his recent unpublished thesis, Barker (2010) has explored the concept of social support with adolescents with CF in order to develop an assessment measure for levels of support. Within this he emphasises the importance of separating out what people do from how this is perceived. That is, supportive behaviours may be helpful on some occasions but not on others depending on the context and perceptions of the person receiving the support (Barker, 2010). Overall, it is likely that both social functioning and social support are pertinent to patients with CF. Future research may wish to further explore their differences and potential impacts on mental health and psychological well-being in CF. In addition, it may be worth considering whether there are differential roles for family members, friends or significant others in providing support or enhancing the social functioning of people with CF.

1.5.2 Quality of reported data

Within reviews it is important to consider the methodological and scientific quality of research papers. Variability in the quality of papers was noted during the review process, although all quantitative papers were felt to be adequately designed. The majority of the studies made use of correlations between multiple factors within their data analyses and research sample numbers

ranged between 16 to 76 participants. A common limitation was in the reporting of statistical power or accounting for multiple comparisons. Only two studies made reference to one of these considerations (Casier, et al., 2011; Riekert, et al., 2007) and not one study reported both the statistical power and use of corrections for multiple statistical comparisons. These two aspects of data analysis have implications for findings and confidence with which conclusions can be drawn. Psychological research commonly aims for a statistical power of .8 and adopts an alpha level of .05 (Barker, Pistrang & Elliot, 2002). When keeping these figures constant, studies with smaller numbers of participants may fail to identify small or medium effects (Barker, Pistrang & Elliot, 2002). Failing to account for multiple comparisons may result in the false identifications of significant results (Kinnear & Gray, 2008). The underreporting of these statistical considerations means that some of the studies identified for review may not be entirely representative of existing relationships.

With regard to the quality of the two qualitative papers, Durst et al. (2001) failed to adequately report important methodological points such as the exact process for data collection, the method used for data analysis and consideration of reliability and validity of the analysis. Qualitative research has the potential to shed light on the lived experience of people with CF, but this needs to be conducted and reported in a manner just as rigorous as quantitative research in order to be viable.

Despite the problems highlighted above in terms of possible statistical weaknesses within quantitative papers, their findings all suggest an inverse relationship between mental health symptoms and social support. It is perhaps the more subtle and finer points of this relationship, such as the influence of moderating variables, which may be misrepresented and therefore need further clarification. Future research needs to involve a greater number of participants and take

account of likely statistical errors resulting from multiple comparisons on potentially underpowered research.

1.5.3 Review limitations

As discussed, most research has limitations and this review is no exception. The current review was aimed to provide an overview of the published research that considered mental health and social support or functioning in adolescents and adults with CF. As a result, it is recognised that those studies or papers which remain unpublished are not represented. The choice to include only published material was based on the availability of time and resources to obtain full-text copies of relevant literature. It may be useful in the future to repeat this review as more papers become published on the topic. A second limitation of this review is found in the fact that only one researcher carried out searches and selection of final papers. The project may have benefited from two or more researchers taking part in every stage of the review process but unfortunately, due to limited resources, this was not possible. Attempts were made to go some way towards minimising selection and assessment bias by involving fellow research colleagues in assessing a random subsection of abstracts for eligibility and collaborating on the standardisation of quality assessment and data extraction forms. Their limited input, however, means that ruling out bias entirely is not possible.

1.6 CONCLUSIONS

Research is beginning to investigate the role of social support for patients with CF and its relationships to mental health symptoms. This review of available published literature points toward an inverse relationship between these two variables. The impact of segregation policies on patients with CF and carers, their coping strategies and experience of mental health symptoms are becoming more frequent topics within the CF literature. The development of

social support measures for people with CF (Barker, 2010) is encouraging and suggests that research within the area may become more prevalent in the coming years.

Although the papers included in this review suggest levels of mental health difficulty as similar to the general population, these studies represent only the literature which includes consideration of social support or functioning. There is a much wider literature on the prevalence of mental health difficulties in CF and a current study is underway to assess this on an international scale (TIDES Project; www.Tides-CF.org). Initial reports from this project are indicating higher levels of mental health difficulty within this population and recommended timing for targeting mental health screening in CF services have been discussed (Goldbeck, et al., 2010; Modi, et al., 2011). Regardless of exact prevalence rates of mental health difficulty in CF, for the subset of CF patients who do experience mental health problems, psychological therapies may be beneficial. Empirical studies assessing the efficacy of psychological therapies are still very much in their infancy (Glasscoe & Quittner, 2008) and benefit cannot automatically be assumed. Traditional cognitive behavioural therapy (CBT) approaches often involve increasing behavioural activities and socialisation. For people with CF, health restrictions, both in terms of physical ability and segregation policies, may impede their opportunity to do this face-to-face. As an alternative to CBT, the recent studies implicating a role for acceptance in aspects of psychological wellbeing mean that acceptance and commitment therapy may have particular value for people with CF. This approach has a growing evidence base spanning a wide range of presenting difficulties (Kangas & McDonald, 2011; Pull, 2009). Future research could have two roles in this area. Firstly, investigating the efficacy of psychological therapies for people with CF and consideration of which components are most active. Secondly, an exploration of how using new technologies (Quittner, et al., 2009) can help overcome socialisation and segregation difficulties.

Research currently underway in the USA is examining the impact of a new web-enabled mobile phone (the CFFone™) on participants' knowledge about CF, social support and treatment adherence (Marciel, Saiman, Quittell, Dawkins, & Quittner, 2010). Although it is unclear whether the researchers also intend to assess mental health or psychological wellbeing, it is possible that by providing this additional means of social support and contact with CF peers, psychological wellbeing may also be improved. In the UK and with regard to the broader spectrum of people living with chronic illness, a recent report has indicated that this population are not receiving adequate emotional and psychological support (Scottish Government and Long Term Conditions Alliance Scotland [LTCAS], 2011). The report recommends the development of services, for example via third sector organisations, to provide greater levels of social support for people with chronic health conditions in order to buffer against later difficulties which may require formal psychological services (Scottish Government & LTCAS, 2011). More specifically to CF, the World Health Organisation's (2002) report recognises the psychosocial challenges faced by patients with CF. It mentions the relationship difficulties which may be encountered throughout life and the detrimental impact that segregation has had on access to peer support (WHO, 2002). To improve social support for CF patients, one possible avenue for exploration is the use of the internet and social networking sites which can offer opportunities for people with CF to interact without the risk of cross-infection. CF-specific discussion forums and online communities provide patients with an online environment for interacting with others in their situation which may help bridge the gap cleaved by the introduction of segregation policies. In addition to the research already being carried out in the USA, it may be useful to investigate the mental health needs of people with CF who already make use of existing internet resources, the ways in which such online forums are used and the types of support they provide.

1.7 REFERENCES

Anderson, D.L., Flume, P.A., & Hardy, K.K. (2001). Psychological functioning of adults with cystic fibrosis. *Chest, 119 (4)*, 1079-1084.

Barker. (2010). *Measurement of disease specific social support in adolescents with cystic fibrosis.* Doctor of Philosophy, University of Miami.

Barker, C., Pistrang, N., & Elliot, R. (2002). *Research methods in clinical psychology: an introduction for students and practitioners* (2nd ed.). Chichester, England.: John Wiley & Sons, Ltd.

Beck, A.T., Steer, R.A., & Barbin, M.G. (1988). Psychometric properties of the Beck Depression Inventory: twenty-five years of evaluation. *Clinical Psychology Review, 8*, 77 - 100.

Bregnballe, V., Thastum, M., & Schiøtz, P.O. (2007). Psychosocial problems in children with cystic fibrosis. *Acta Pædiatrica, 96*(1), 58-61. doi: 10.1111/j.1651-2227.2006.00014.x

Burker, E.J., Carels, R.A., Thompson, L.F., Rodgers, L., & Egan, T. (2000). Quality of life in patients awaiting lung transplant: Cystic fibrosis versus other end-stage lung diseases. *Pediatric Pulmonology, 30 (6)*, 453-460.

Burker, E.J., Sedway, J., Carone, S., Burker, E.J., Sedway, J., & Carone, S. (2004). Psychological and educational factors: better predictors of work status than FEV1 in adults with cystic fibrosis. [Research Support, Non-U.S. Gov't]. *Pediatric Pulmonology, 38*(5), 413-418.

Casier. (2011). Acceptance and well-being in adolescents and young adults with CF. *Journal of Pediatric Psychology*, *36* (4), 476 - 489.

Cella, D. (1998). Quality of life. In J. C. Holland (Ed.), *Psychooncology* (pp. 1135 - 1146). New York, Oxford: Oxford University Press.

Cohen, S., & Wills, T. (1985). Stress, social support, and the buffering hypothesis. *Psychological Bulletin, 98*(2), 310-357.

Coyne, J.C., & Downey, G. (1991). Social Factors and Psychopathology: Stress, Social Support, and Coping Processes. *Annual Review of Psychology, 42*(1), 401-425. doi: doi:10.1146/annurev.ps.42.020191.002153

Cystic Fibrosis Trust (2011). *About cystic fibrosis.* Retrieved 3rd March 2011, from www.cftrust.org.uk/aboutcf

Delelis, G., Christophe, V., Leroy, S., Vanneste, J., & Wallaert, B. (2008). The effects of cystic fibrosis on couples: Marital satisfaction, emotions, and coping strategies. *Scandinavian Journal of Psychology, 49 (6)*, 583-589.

Durst, C.L., Horn, M.V., MacLaughlin, E.F., Bowman, C.M., Starnes, V.A., & Woo, M.S. (2001). Psychosocial responses of adolescent cystic fibrosis patients to lung transplantation. *Pediatric Transplantation, 5*(1), 27-31.

Ernst, M.M., Johnson, M.C., & Stark, L.J. (2010). Developmental and psychosocial issues in cystic fibrosis. *Child & Adolescent Psychiatric Clinics of North America, 19*, 263 - 283.

Forgeron, P.A., King, S., Stinson, J.N., McGrath, P.J., MacDonald, A.J., & Chambers, C.T. (2010). Social functioning and peer relationships in children and adolesents with chronic pain: A systematic review. *Pain Research and Management: The Journal of the Canadian Pain Society, 15*(1), 27 - 41.

Geddes, D. (2001). Of isoates and isolation: Pseudomonas aeruginosa in adults with cystic fibrosis. *Lancet, 358*, 522 - 523.

Glare, P., Pereira, G., Kristjanson, C. J., Stockler, M., & Tattersall, M. (2004). Systematic review of the efficacy of antiemetics in the treatment of nausea in patients with far-advanced cancer. *Support Care Cancer, 12*, 432 - 440.

Glasscoe, C. A., & Quittner, A. L. (2008). Psychological interventions for people with cystic fibrosis and their familes. *Cochrane Databse of Systematic Reviews*, Art. No.: CD003148.

Goldbeck, L., Besier, T., Hinz, A., Singer, S., Quittner, A.L., & Group, T. (2010). Depression in German patients with cystic fibrosis. *Chest, 138*(4), 929 - 936.

Havermans, T., Colpaert, K., & Dupont, L.J. (2008). Quality of life in patients with Cystic Fibrosis: Association with anxiety and depression. *Journal of Cystic Fibrosis, 7*(6), 581-584.

Hefner, J., & Eisenberg, D. (2009). Social Support and Mental Health Among College Students. *American Journal of Orthopsychiatry, 79*(4), 491-499. doi: 10.1037/a0016918

Herzer, M., Umfress, K., Aljadeff, G., Ghai, K., & Zakowski, S.G. (2009). Interactions with parents and friends among chronically Ill children: Examining social networks. *Journal of Developmental and Behavioral Pediatrics, 30 (6)*, 499-508.

Holahan, C.J., & Moos, R.H. (1981). Social support and psychological distress: a longitudinal analysis. *Journal of Abnormal Psychology, 90*(4), 365 - 370.

Ishihara, L., & Brayne, C. (2005). A systematic review of nutritional risk factors in Parkinson's disease. *Nutritional Research Reviews, 18*, 259 - 282.

Kangas, M., & McDonald, S. (2011). Is it time to act? The potential of acceptance and commitment therapy for psychological problems following acquired brain injury. *Neuropsychological Rehabilitation, 21*, 250 - 276.

Kinnear, P.R., & Gray, C.D. (2008). *SPSS 15 made simple*. East Sussex, England: Psychology Press.

Liang, H. Y., Zhou, H., Li, X. L., Yin, Z. H., Guan, P., & Zhou, B. S. (2010). Chemo-radiotherapy for advanced non-small cell lung cancer: concurrent or sequential? It's no longer the question: a systematic review. *International Journal of Cancer, 127* (3), 718 - 728.

Liddle, J., Williamson, M., & Irwig, L. (1996). *Method for evaluating research and guideline evidence*. Sydney: New South Wales Department of Health.

Littlewood, J. (2004). *Looking back over 40 years and what the future holds.* Paper presented at the 27th European Cystic Fibrosis Conference, Birmingham.

Macdonald, K. (2006). Living in limbo--patients with cystic fibrosis waiting for transplant. *British journal of nursing (Mark Allen Publishing), 15 (10)*, 566-572.

Marciel, K.K., Saiman, L., Quittell, L.M., Dawkins, K., & Quittner, A.L. (2010). Cell phone intervention to improve adherence: Cystic fibrosis care team, patient, and parent perspectives. *Pediatric Pulmonology, 45*(2), 157-164. doi: 10.1002/ppul.21164

Mann, M., Hosman, C.M.H., Schaalma, H.P., & de Vries, N.K. (2004). Self-esteem in a broad-spectrum approach for mental health promotion. *Health Education Research, 19*(4), 357-372. doi: 10.1093/her/cyg041

Modi, A.C., Driscoll, K.A., Montag-Leifling, K., & Acton, J.D. (2011). Screening for symptoms of depression and anxiety in adolescents and young adults with cystic fibrosis. *Pediatric Pulmonology, 46*(2), 153-159. doi: 10.1002/ppul.21334

Pearson, D.A., Pumariega, A.J., & Seilheimer, D.K. (1991). The Development of Psychiatric Symptomatology in Patients with Cystic Fibrosis. *Journal of the American Academy of Child and Adolescent Psychiatry, 30*(2), 290-297.

Pfeffer, P.E., Pfeffer, J.M., & Hodson, M.E. (2003). The psychosocial and psychiatric side of cystic fibrosis in adolescents and adults. [Review]. *Journal of Cystic Fibrosis, 2*(2), 61-68.

Public Health Resources Unit (2006). *10 questions to help you make sense of qualitative research*. England.

Pull, C. B. (2009). Current empirical status of acceptance and commitment therapy. *Current Opinion in Psychiatry, 22,* 55-60 10.1097/YCO.1090b1013e32831a32836e32839d.

Quittner, A.L., Barker, D., Marciel, K., & Grimley, M.E. (2009). Cystic fibrosis: A model for drug discovery and patient care. In M. C. Roberts & R. G. Steele (Eds.), *Handbook of Pediatric Psychology* (4th ed.): Guilford Press.

Quittner, A.L., Barker, D.H., Snell, C., Grimley, M.E., Marciel, K., & Cruz, I. (2008). Prevalence and impact of depression in cystic fibrosis. *Current Opinion in Pulmonary Medicine, 14*(6), 582-588 510.1097/MCP.1090b1013e3283121cf3283121.

Quittner, A.L., Buu, A., Watrous, M., & Davis, M.A. (2002). Cystic Fibrosis Questionnaire - Revised: Forest Laboratories.

Riekert, K.A., Bartlett, S.J., Boyle, M.P., Krishnan, J.A., & Rand, C.S. (2007). The association between depression, lung function, and health-related quality of life among adults with cystic fibrosis. *Chest, 132*(1), 231 - 237.

Russo, K., Donnelly, M., & Reid, A.J.M. (2006). Segregation--the perspectives of young patients and their parents. *Journal of Cystic Fibrosis, 5*(2), 93-99.

Scottish Government & Long Term Conditions Alliance Scotland (2011). Emotional support matters: emotional and psychological support needs of people with long term conditions. Glasgow: Scottish Government.Last retrieved on 16th May 2011 from http://www.ltcas.org.uk/documents/LTCASEmotionalReportFinal.pdf.

Scottish Intercollegiate Guidelines Network (2008). SIGN 50: A guideline developer's handbook. (Revised ed.): SIGN, Edinburgh.

Singleton, N., Bumpstead, R., O'Brien, M., Lee, A., & Meltzer, H. (2001). *Psychiatric morbidity among adults living in private households, 2000.* London: HMSO.

Soon, Y. Y., Stockler, M., Askie, L. M., & Bajer, M. J. (2009). Duration of chemotherapy for advanced non-small-cell lung cancer: a systematic review and meta-analysis of randomized trials. *Journal of Clinical Oncology, 27* (20), 3277 - 3283.

Spilker, B. (1996). Introduction. In B. Spilker (Ed.), *Quality of life and pharmacoeconomics in clinical trials* (pp. 1 - 10). Philadelphia, PA: Lippincott-Raven.

Szyndler, J.E., Towns, S.J., van Asperen, P.P., & McKay, K.O. (2005). Psychological and family functioning and quality of life in adolescents with cystic fibrosis. *Journal of Cystic Fibrosis, 4 (2)*, 135-144.

White, T., Miller, J., Smith, G.L., McMahon, W.M., White, T., Miller, J. (2009). Adherence and psychopathology in children and adolescents with cystic fibrosis. *European Child & Adolescent Psychiatry, 18*(2), 96-104.

World Health Organisation (2002). Services for adults with cystic fibrosis: report of a joint WHO/ICF(M)A/IACFA meeting. WHO. Last retrieved on 3rd October 2011 from http://www.cfww.org/docs/who/2002/services_for_adults_with_cystic_fibrosis.pdf.

Zigmund, A.S., & Snaith, R.P. (1983). The Hospital Anxiety and Depression scale. *Acta Psychiatr Scand, 67*, 361 - 370.

2 THESIS AIMS

With the vast majority of people with cystic fibrosis (CF) now living into adulthood researchers have become increasingly interested in their long-term quality of life and psychological well-being. Studies have indicated that the CF population report high levels of quality of life (Abbott, 2009) but that levels of mental health difficulty vary. Some studies suggest higher levels of depression and/or anxiety compared to the general population (Goldbeck *et al.*, 2010; Modi *et al.*, 2011; Quittner *et al.*, 2008) whereas others maintain that levels of mental health symptoms are comparable to the general population (Anderson *et al.*, 2001). The reasons for this variability continue to be explored by research and factors identified as influential include being female, older, unemployed and having more severe illness (Goldbeck *et al.*, 2010; Modi *et al.*, 2011; Quittner *et al.*, 2008).

Two factors which have been shown to play an important role in the mental health of the general population are self-esteem and social support. However, these two variables have received relatively little attention within the CF literature. Self-esteem has been linked with better mental health and psychological wellbeing in the general population (Mann *et al.*, 2004) and has been shown to play an important role in mediating the relationship between dysfunctional thoughts and depression in adult participants (Simpson *et al.*, 2010). To the authors' knowledge, only two papers have explicitly assessed self-esteem in adults with CF with both papers focusing primarily on body image (Abbott *et al.*, 2000; Abbott *et al.*, 2007). A recent publication by Brucefors *et al.* (10) speculated that low levels of self-esteem may have contributed to the higher levels of social dysfunction found in their sample of adults with CF compared with healthy adults. However, there was no direct measurement of self-esteem within their project and, as yet,

there appear to be no published studies which consider the relationship between self-esteem and mental health in adults with CF.

Social support is important for adaptation to stress and higher levels of support are related to lower levels of mental health difficulty in the general population (Hefner & Eisenberg, 2009). Peer relationships have been seen to be affected in chronic illnesses (Forgeron *et al.*, 2010) and, with the use of segregation in CF services to prevent cross-infection of respiratory bacteria, it is important to consider how people with CF can build and maintain relationships with and access support from their CF peers (Quittner *et al.*, 2009). The evidence from CF research overall seems to indicate that higher levels of social support and better levels of social functioning are related to better coping (White *et al.*, 2009), improved adherence to treatment (White *et al.*, 2009) and fewer mental health difficulties (Casier *et al.*, 2011). Researchers have suggested that future studies continue to examine the potential buffering effects of social support (Modi *et al.*, 2011) and explore how new technologies may enhance peer interaction and overcome the restrictions of segregation for people with CF (Quittner *et al.*, 2009).

One option for keeping in contact with CF peers is to use the internet and social networking websites, where interactions can take place without the risks of cross-infection. Online resources are becoming increasingly popular (Hasenecz, 2010) and may provide an additional means of peer support for people with CF. However, little research has been carried out in the area of online resources, the characteristics of CF patients who make use of specific resources and the topics of discussion that arise.

This thesis presents two studies aimed at adding to the existing literature on self-esteem in adults with CF and providing an exploration of how a CF-specific online discussion forum is used.

Study 1 consisted of a quantitative survey including standardised measures of mental health symptoms, self-esteem and HRQoL. Study 2 took a qualitative approach to explore the topics of discussion which arose within an online discussion forum hosted by a UK-based charity, the Cystic Fibrosis Trust (CF Trust).

2.1 STUDY 1 - RESEARCH QUESTIONS

The first study aimed to answer two primary research questions:

a) What are the levels of mental health symptoms, HRQoL and self-esteem in adults with CF who make use of online discussion forums and internet resources?

b) Are there significant correlations between measures of HRQoL, mental health and self-esteem in adults with CF?

Levels of mental health difficulty were hypothesised to be similar to recent research in CF which has indicated a slight elevation in the prevalence of mental health difficulty when compared to the general population. In line with previous research involving the general population and CF patients, it was hypothesised that lower self-esteem would be related to poorer mental health and lower levels of quality of life.

2.2 STUDY 2 - RESEARCH QUESTION

The second study aimed to answer the following question:

a) What topics are discussed and what types of support are sought from a CF-specific online discussion forum?

2.3 ETHICAL CONSIDERATIONS

Ethical approval for the two studies was sought and obtained from the School of Health in Social Science, University of Edinburgh research ethics panel (Appendix B). Collaboration with the Cystic Fibrosis Trust aided the process of refinement for the project to ensure that participants

were able to give fully informed consent. An information page posted on the CF Trust website provided details on how data would be used and stated that participants were able to withdraw at any time without giving a reason. Data were kept anonymous and participants chose whether to provide their online discussion forum username. The CF Trust provides guidance on using their online forum and advises users not to provide any identifying personal information. Forum usernames were only used to identify discussion board posts to be included in qualitative analysis. Participants were informed that any direct quotes used within the report would not include the username of its author. At the end of the online survey, a number of support contacts were included and participants were encouraged to speak with a member of their health team if any items within the standardised measures had raised issues for them.

3 STUDY 1 METHODS

3.1 DESIGN

This was a one time-point observational study of a population of adults with cystic fibrosis who made use of online discussion forums and social networking. Participants completed three quantitative assessment measures and provided demographic information.

3.2 PARTICIPANTS

Participants were recruited through an online discussion forum and Facebook social networking page hosted by the Cystic Fibrosis Trust (CF Trust), a UK-based charity. A total of 105 eligible participants consented to take part in the study. Surveys were completed to varying levels with fully completed data sets submitted by 74 participants (see Table 3.1).

Table 3.1 Number of survey sections completed out of 105 consenting participants

Survey Section	Number (%) completed	
Demographic information	89 (84.8)	
CFQ-R	78 (74.3)	Full sets of data
CORE-OM 34	75 (71.4)	N = 74 (70.1%)
Rosenberg self-esteem	75 (71.4)	
Username only	5 (4.8)	
No information given	11 (10.5)	

Key demographic details for full and partially completed surveys are presented in Table 3.2. Non-parametric comparisons of key characteristics of full- and partial- completers were carried out using chi-squared and Mann-Whitney U tests. There were no significant differences between groups on any variable: age (U = 383.5; exact p = .14, n.s.); FEV_1% predicted (U = 294.0; exact p = .69, n.s.); gender (x^2 = 3.56; exact p = .11, n.s.); ethnicity (x^2

= 0.60; exact p = 1.00, n.s.); country (x^2 = 0.20; exact p = 1.00, n.s.); lung transplant (x^2 = 0.02; exact p = 1.00, n.s.). These results indicate that the two groups of participants did not differ significantly in terms of demographic variables. A more detailed breakdown of demographic variables for those participants completing a full set of survey data are presented in Table 3.3.

Table 3.2 Key demographic characteristics of full and partial surveys completed

	Full	**Partial**
N	74	15
Age (mean ± SD)	27.8 ± 9.2	31.0 ± 8.7
Gender (% female)	77.0%	53.3%
Ethnicity (% White)	97.3%	93.3%
Country (% UK/Ireland)	96.0%	93.3%
Education (% Higher Ed)	60.8%	53.3%
FEV_1% (mean ± SD)	61.7 ± 25.0	58.0 ± 21.0
Lung transplant (% yes)	6.8%	7.7%

3.3 PROCEDURE

Recruitment was initiated via a message posted on the CF Trust discussion forum by a member of the CF Trust communications team. This linked to an information page describing the study to forum users. Notices were placed within the 'teenage' and 'adult' sections of the discussion forum. After 3 and 6 weeks, the researcher replied to the initial post to thank those who had already taken part, to encourage further participation and also to bring the notice to the top of the discussion area. In addition, 6 weeks after the original post on the CF Trust discussion forum, the communications team placed a recruitment notice

Table 3.3 Demographic and basic clinical information summary for full data sets

Demographic	**N (%)**	**Clinical**	**N (%)**
Marital Status (N = 74)		Age at CF diagnosis (N = 74)	
Single/never married	28 (37.8)	*At birth*	19 (25.7)
With a partner	25 (33.8)	*<6 months old*	18 (24.3)
Married/civil partnership	20 (27.0)	*6 – 12 months old*	10 (13.5)
Widowed	1 (1.4)	*1 – 4 years old*	13 (17.5)
		5 – 11 years old	3 (4.1)
Education (N = 74)		*12 – 17 years old*	3 (4.1)
Some secondary school or less	1 (1.3)	*18 – 25 years old*	3 (4.1)
GCSEs/O-levels	11 (14.9)	*>25 years old*	5 (6.7)
A/AS-levels	17 (22.9)		
Other higher education	13 (17.6)	CF Severity, No Transplant (FEV_1%; N = 54)	
University degree	21 (28.4)	*Normal (FEV_1% ≥90%)*	6 (11.1)
Professional or postgraduate	11 (14.9)	*Mild (70 – 89%)*	13 (24.1)
		Moderate (40 - 69%)	21 (38.9)
Work Status (N = 61)		*Severe (<40%)*	14 (25.9)
Working full- or part-time	32 (52.5)		
Not attending school or work due to health	12 (19.7)	CF Severity, Transplant (FEV_1%; N = 4)	
Attending school	11 (18.0)	*Normal (FEV_1% ≥90%)*	3 (75.0)
Seeking work	2 (3.3)	*Moderate (40 - 69%)*	1 (25.0)
Not working for other reasons	2 (3.3)		
Taking education courses at home	1 (1.6)		
Full-time homemaker	1 (1.6)		

on the CF Trust Facebook page followed by a repeat post from the researcher 2 weeks later. In total, the online survey was open for ten weeks.

The information page contained a web-link to the online survey, hosted by Survey Monkey (www.surveymonkey.com). Participants self-selected themselves to take part in the research and were eligible if they were 16 years old or over and had a diagnosis of cystic fibrosis. Participants were asked to provide consent prior to completing the online survey. Those who chose not to consent were directed to the end of the survey and thanked for their time.

3.4 MEASURES

3.4.1 Cystic Fibrosis Questionnaire – Revised (CFQ-R)

The CFQ-R (Bryon & Stramik, 2005; Quittner *et al.*, 2002) is a measure of health-related quality of life (HRQoL) designed especially for people diagnosed with CF. The UK teen/adult version (for people aged 14 years old and above) was used in this instance. The measure consists of 50 items which assess HRQoL within 9 domains: physical functioning, health perceptions, vitality, respiratory symptoms, treatment burden, role functioning, emotional functioning and social functioning. The CFQ-R does not result in a single composite score. Previous research advocates for the consideration of four core domains when studying quality of life (Casier *et al.*, 2011; Cella, 1998; Spilker, 1996), as a result, it was decided to include the CFQ-R subscales of physical functioning (eight items; e.g. 'I have trouble recovering after physical effort'), emotional functioning (five items; e.g. 'I often feel lonely'), role functioning (four items; e.g. 'How often does CF get in the way of meeting your school, work, or personal goals?') and social functioning (six items; e.g. 'I get together with my friends a lot'). Each item on the CFQ-R is rated on a four point Likert scale. A standardised score ranging between 0 – 100 is obtained for each of the subscales, with higher scores indicating better functioning. The CFQ-R teen/adult version has been

shown to be well established, valid and reliable (Palermo *et al.*, 2008). Cronbach's alpha internal consistency analyses were carried out for each of the four subscales: physical α = .96, emotional α = .82, role α = .86 and social α = .63.

3.4.2 Clinical Outcomes in Routine Evaluation – Outcome Measure 34

The CORE-OM (Evans *et al.*, 2000) is a self-report measure consisting of 34 items to assess psychological and emotional distress in terms of four sub-domains: subjective well-being, functioning, symptoms and risk. A total score is also calculated and is used throughout this study. Each item is rated on a four-point scale from 0 (not at all) to 4 (most or all of the time), with higher scores indicating greater difficulties. The measure has been shown to be reliable, valid and able to distinguish between clinical and non-clinical samples in the general population (Evans *et al.*, 2002), primary care settings (Gilbody & Barkham, 2007) and patients with type 1 diabetes (Masding *et al.*, 2011). In this instance, internal consistency tests of reliability revealed Cronbach's alpha of .96.

3.4.3 Rosenberg self-esteem scale (Rosenberg, 1965)

This 10-item brief self-report questionnaire is designed to measure levels of global self-esteem in adolescents and adults. Each item is rated on a four-point scale (strongly disagree, disagree, agree, strongly agree) and produces a total score ranging from 10 – 40. Higher scores represent higher levels of self-esteem. This measure of self-esteem has been used in chronic illness populations including samples of adults with CF (Abbott *et al.*, 2000; Abbott *et al.*, 2007; Ireys *et al.*, 1994) and has been shown to have satisfactory psychometric properties (Sinclair *et al.*, 2010). Internal consistency tests of reliability revealed a Cronbach's alpha of .94.

3.4.4 Demographic questionnaire

General information was collected using a set of questions developed by the researcher in collaboration with two health colleagues. This included age, gender, occupation, diagnosis,

current physical health status (including most recent FEV_1% predicted reading and lung transplant status) and frequency of discussion forum use. FEV_1% predicted is a measure of lung function frequently used to classify severity of respiratory disease. Participants were asked how often they made use of the CF Trust discussion forum and/or Facebook page. They also indicated from a list of options, the things they used each site for. Where participants indicated that they used the CF Trust discussion forum, two open-ended questions asked what users liked most and least about the forum.

3.5 DATA ANALYSIS

3.5.1 Quantitative data

Raw data were entered into SPSS version 17 where coding and total scores were calculated for each assessment measure. Scores for those participants who had fully completed the online survey (N = 74) were included in analyses. Kolmogorov-Smirnov statistical tests were carried out to determine the normality of data distribution for FEV_1% predicted and each outcome measure variable. Normal distributions of data were found for FEV_1% predicted ($D = .95$, $p = .67$); CORE total ($D = .11$, $p = .32$); Rosenberg self-esteem scale ($D = .12$, $p = .26$) and CFQ-R subscales of emotional ($D = .11$, $p = .38$), social ($D = .09$, $p = .60$) and role ($D = .14$, $p = .11$) functioning. The CFQ subscale scores for physical functioning were not normally distributed ($D = .17$, $p = .03$)[1].

[1] The researcher explored the benefit of transforming data using a square-root function. This succeeded in distributing scores normally ($D = 0.14$, $p = .11$). However, as the transformed scores did not result in any differences in the significance of subsequent statistical analyses compared to when using raw scores, it was decided to use the raw data for ease of interpretation of findings.

3.5.1.1 Correlations and power analysis

Correlations using Pearson product moment analyses were conducted between the following variables: physical health status (as measured by self-reported FEV_1% predicted); CFQ-R physical, social, role and emotional functioning; CORE-OM total score; and Rosenberg self-esteem scale total score. Consideration was given to removing data of those participants who had undergone a lung transplant prior to statistical analysis. Given the small number of participants for whom this was applicable (N = 5) and the fact that mean scores were not changed significantly when they were excluded, it was decided to include their data in subsequent analyses.

Calculations were carried out using G*Power version 3.1.2 to establish the size of correlation which could be confidently detected for a sample size of 74, an alpha level of .05 and statistical power of 0.8. With a two-tailed hypothesis this revealed analysis could detect correlations of r = .31, a medium effect size (Cohen, 1988).

3.5.1.2 Gender comparison

Past research has suggested gender differences in quality of life and psychological well-being scores in CF populations (e.g. Anderson, *et al.*, 2001; Gee *et al.*, 2003). In light of the unequal split of males and females within the study sample it was felt important to explore whether the overrepresentation of females had impacted on the mean levels of wellbeing and HRQoL in the present study. As a result, t-test comparisons of male and female mean scores on each outcome measure and FEV_1% predicted were carried out. It was recognised that due to the small sample, these comparisons were unlikely to meet statistical power to detect small or medium effect sizes. However, it was felt that exploring this area of potential influence on data remained important in order to interpret findings and consider them against existing studies which include an equal split of males and females.

3.5.2 Online resource questions

Data indicating how frequently participants used the CF Trust discussion forum and Facebook page were summarised using percentages. For the two open-ended questions, responses were collated and summarised on the basis of themes mentioned across the data set. Analysis of these questions was carried out for all available data whether or not standardised measures had been fully completed.

4 STUDY 1 RESULTS

4.1 DESCRIPTIVE STATISTICS

Table 4.1 presents the mean and standard deviation (SD) for each measure completed within the online survey.

It can be seen that average scores on the CFQ-R subscales vary. The lowest score was found for physical functioning (mean = 46.96, SD = 32.50) and the highest for role functioning (mean = 65.61, SD = 23.40). Mean scores of 57.30 (SD = 24.62) and 59.23 (SD = 20.38) were obtained for emotional and social functioning respectively. Self-esteem scores from the online survey produced a mean score of 29.89 (SD = 7.08). The mean score on the CORE-OM 34 was 1.01 (SD = 0.72) and, when classified according to clinical thresholds, 29.7 per cent (N = 22) of participants reported clinical levels of symptoms.

Table 4.1 Means and standard deviations of outcome measure scores by gender

Outcome measures		**Mean scores (SD)**		
		Male	Female	Total
FEV_1% predicted		66.73 (26.9)	60.47 (24.6)	61.66 (25.0)
CFQ-R	*Physical functioning*	58.82 (37.6)	43.42 (30.3)	46.96 (32.5)
	Role functioning	71.57 (23.8)	63.73 (23.2)	65.61 (23.4)
	*Emotional functioning***	71.76 (20.9)	52.98 (24.2)	57.30 (24.6)
	*Social functioning***	70.59 (15.6)	55.85 (20.5)	59.23 (20.4)
CORE-OM total score*		0.64 (0.5)	1.13 (0.7)	1.01 (0.7)
Rosenberg self-esteem scale**		33.94 (5.1)	28.68 (7.2)	29.89 (7.1)

** gender difference significant at the level of p<.05*

*** gender difference significant at the level of p<.01*

4.2 CORRELATIONS

Pearson product moment analyses were conducted to establish whether relationships existed between variables of disease severity (as measured by FEV_1% predicted), HRQoL, mental health and self-esteem. The results of the correlation analyses are presented in Table 4.2. As would be expected, significant positive correlations were found between the four subscales of the CFQ-R.

Correlations between CFQ-R subscales and CORE-OM total score were all negative, with lower quality of life scores associated with higher levels of mental health difficulty. Significant positive correlations between CFQ-R subscales and self-esteem scores were found, indicating that higher levels of self-esteem are related to better HRQoL. Higher self-esteem scores were also associated with lower levels of mental health difficulty as shown by the negative correlation between the CORE-OM total and Rosenberg self-esteem scale scores. Lung functioning was seen to correlate positively with CFQ-R subscales of physical, role and social functioning. This suggests that those with better lung function experience better quality of life in these domains. The only non-significant correlations were found for disease severity with emotional functioning, CORE-OM total score and self-esteem.

Table 4.2 Correlation coefficients and significance for outcome measures

		1	2	3	4	5	6	7
1. FEV_1% Predicted	r	1						
	N	58						
2. Physical functioning	r	.759*	1					
	N	58	74					
3. Role functioning	r	.445*	.757*	1				
	N	55	71	71				
4. Emotional functioning	r	.232	.569*	.686*	1			
	N	58	74	71	74			
5. Social functioning	r	.517*	.722*	.713*	.632*	1		
	N	58	74	71	74	74		
6. CORE-OM total score	r	-.135	-.464*	-.587*	-.841*	-.528*	1	
	N	58	74	71	74	74	74	
7. Rosenberg Self-esteem score	r	.233	.482*	.609*	.733*	.589*	-.827*	1
	N	58	74	71	74	74	74	74

** Correlation is significant at the p<.01 level (two-tailed)*

4.3 GENDER DIFFERENCES

Exploratory two-tailed t-tests were carried out to compare scores of males (N = 17) and females (N = 57) on each of the outcome measures. Mean scores, standard deviations and significance can be found in Table 4.1. Females were found to score significantly lower than males on the CFQ-R domains of emotional functioning (t = 2.897, p = .005) and social functioning (t = 2.730; p = .008). Females were also seen to have lower levels of self-esteem (t = 2.813; p = .006) and higher total CORE-OM scores of mental health symptoms (t = 2.534; p = .013). No significant gender differences in FEV_1% predicted (t = 0.746; n.s.), physical functioning (t = 1.739; n.s.) or role functioning (t = 1.208; n.s.) were found.

4.4 USE OF ONLINE RESOURCES

The frequency with which participants made use of the CF Trust discussion forum and Facebook page varied (N = 85). Half of the participants (N = 44, 51.8 per cent) used only the discussion forum, 24.7 per cent (N = 21) used only the Facebook page, whilst a further 23.5 per cent (N = 20) used both online resources. Frequency of visits to and posts made on the CF Trust discussion forum varied. Data were available for all but one of the participants who used the CF Trust forum (N = 63). Most forum users visited the site either everyday (N = 16, 25 per cent) or 3 to 4 times per week (N = 28, 43.8 per cent). Smaller numbers of participants visited the site once a week (N = 7, 10.9 per cent), 2 to 3 times per month (N = 8, 12.5 per cent) and once a month or less (N = 4, 6.3 per cent). Much less frequent were the amount of posts made on the discussion forums. The majority of users posted once a month or less (N = 25, 39.1 per cent), whilst fewer participants posted 2 to 3 times a month (N = 8, 12.5 per cent), once a week (N = 9, 14.1 per cent), 3 to 4 times a week (N = 8, 12.5 per cent) and everyday (N = 3, 4.7 per cent). A proportion of forum users never posted on the site and chose to only read posts (N = 10, 15.6 per cent).

For those participants indicating usage of the discussion forum or Facebook page, indication was given as to the purposes for which these sites were used. These are presented in Table 4.3. Seven participants also provided comments on their use of the online resources not included within the survey options; five people said they use the sites to help others and offer advice, one person signed up to support the site and one person used the site to find out about television appearances by people with CF.

Table 4.3 Uses of discussion forum (N = 64) and Facebook page (N = 40)

	CF Trust discussion forum	**CF Trust Facebook page**
Find information about CF	62 (96.9%)	20 (50.0%)
Find information about CF treatments	56 (87.5%)	13 (32.5%)
Get health advice	48 (75%)	14 (35.5%)
Pass the time when in hospital	26 (40.6%)	19 (47.5%)
Keep in touch with CF friends	22 (34.4%)	15 (37.5%)
Get advice about things other than CF	17 (26.6%)	10 (25.0%)
Meet new people	15 (23.4%)	15 (37.5%)
Find out about events	14 (21.9%)	18 (45.5%)
Arrange fundraising events	11 (17.2%)	6 (15.0%)

4.4.1 Open-ended questions

Participants who indicated use of the CF Trust discussion forum were asked to comment on the things they liked a) most and b) least about the forum. A total of 62 and 56 participants, respectively, chose to comment. Themes of these comments are described below.

4.4.1.1 Things most liked about the CF Trust discussion forum (N = 62)

Some aspects of the forum which people liked most were the shared experiences and understanding between forum members and the support that people offered on the site. Many people commented that they liked the availability of advice, information and opportunity to discuss CF generally and, more specifically, others' experiences of treatment. A number of people commented that they liked meeting new people and that the forum helped to overcome the restrictions resulting from segregation policies. In addition, some users mentioned that they felt reassured that they were not alone in coping with CF and they valued having contact with other people who were 'in the same boat'. A few participants mentioned that they felt the forum had a friendly atmosphere where people were honest about their opinions and feelings. Others appreciated the range of experiences that members had and felt there was a sense of community between the forum members. Finally, it was also mentioned that the site was well-monitored by the CF Trust moderators.

4.4.1.2 Things liked least about the CF Trust discussion forum (N = 56)

By far the most common feature which was liked least about the discussion forum was the occurrence of arguments and disagreements amongst members. Many people commented on this along with the occasional insensitivity, personal insults and lack of respect within some forum posts. However, a number of participants commented that these issues arose in everyday life and were a normal part of relationships. Others commented that they disliked the 'cliques' within the forum user group. Another common comment made by participants was the tendency for the forum to be negative in terms of some content and information which led people to feel lower in mood. This was highlighted in terms of posts being a reminder of the daily difficulties faced when coping with CF, the future progression of the disease and notifications of people who had died. A minority of participants commented that they felt the relevance of some topics should be more closely related to CF, that people often asked questions previously discussed on the board and that getting replies to posts can be

slower than other social networking websites (such as Facebook). A number of comments related less to the content of posts and more to the organisation of the discussion forum in general. Some felt that the moderation by the CF Trust could inhibit discussion or be heavy-handed. Others would prefer if the forum was split into more defined discussion areas, that users had the option to send personal messages (not publicly visible) to others and that the forum was updated in its appearance.

4.5 SUMMARY

The results indicate that the participants have variable levels of HRQoL depending on domain. Lowest scores were obtained in physical functioning whilst highest functioning was seen in relation to roles. The sample seemed to have good levels of self-esteem despite their poor levels of physical functioning. Mental health symptoms were at clinical levels for 29.7 per cent of the sample, with an average total score on the CORE-0M of 1.01. Significant correlations were revealed between measures of mental health, self-esteem and HRQoL indicating that better levels of HRQoL are related to higher levels of self-esteem and lower levels of mental health difficulty. Furthermore, females were seen to experience lower levels of emotional and social functioning, poorer self-esteem and higher levels of mental health difficulty compared to males.

In relation to the use of online resources, participants used the CF Trust discussion forum and Facebook page to differing degrees with the majority of people visiting the discussion forum more than twice per week. Posting on the forum, however, was much less frequent, with most people posting only once a month. It seems that the two online resources provide CF patients with the opportunity to interact with and meet other people with CF without risks of cross-infection. In addition, the availability of information, advice, shared understanding and support appear to be valued aspects of the forum. Participants acknowledged that there could be difficulties in interactions similar to those which play out in non-virtual relationship

such as arguments and misunderstandings. Despite this, participants still reported a sense of community, support and solidarity with their fellow forum-users.

This study has provided an insight into the levels of mental health difficulty, self-esteem and quality of life within this population along with their views on elements of the discussion forum which they like or dislike. The second study aims to explore the range of topics discussed and the way the forum is used through a qualitative thematic analysis of discussion forum posts.

5 STUDY 2 METHODS

5.1 DESIGN

This study aimed to systematically examine discussion posts made on an online discussion forum aimed at people whose lives are affected by CF. Qualitative data were collected from the forum retrospectively using a time period of one month.

5.2 PROCEDURE

Participants were recruited to take part in the study via Study 1. Usernames provided by participants were gathered and the first 25 users consenting to take part in the study were identified. All participants providing their username had consented for posts made over the preceding six months to be used in the research. However, due to the high response rate, it was decided to take a sample of data from the first 25 participants' posts over a 4 week period. Posts made during the month preceding the first recruitment notice were collected and organised by author in chronological order. Eight of the 25 users had made no posts during the time period specified. Forum posts from the remaining 17 users were entered into a word processing document for thematic analysis. The final sample of forum data used in this study consisted of 404 posts across 171 discussion threads made by 17 different forum users.

5.3 PARTICIPANTS

A total of 55 participants supplied their CF Trust forum username to allow research to collect their forum posts for analysis. All participants were adults over the age of 16 years who had a diagnosis of CF. For the 17 participants whose data were used, their average age was 29.9 years (SD = 7.8, range 20 to 48 years). The majority of users were female (N = 12, 70.6 per cent) and 2 (11.8 per cent) were known to have had a lung transplant. On average, the forum users had been a member of the CF Trust for 3.1 years (SD = 1.3, range 0.08 – 4.2 years).

5.4 DISCUSSION FORUM

At the time of recruitment, the CF Trust discussion forum had a total of 4649 registered members and consisted of six discussion areas: CF teenagers, CF adults, parents and carers, partners of people with CF, fundraising forum and runners' forum. The forum is open to the public and visitors to the site need not register in order to read threads. Within each discussion area, registered users may start a new topic of conversation by posting a comment or question. Other users may then respond with their own contributions, forming a discussion or 'thread'. The CF Trust provide 'house rules' informing users of good practice on the discussion boards, including advising against disclosure of personally identifiable information, avoiding offensive posts and advertising. A group of administration staff and moderators oversee the use of the forum to ensure appropriate postings. They are able to lock threads to prevent further discussion on a topic or delete those which are inappropriate for the forum (e.g. spam messages or those deemed offensive).

5.5 DATA ANALYSIS

5.5.1 Qualitative data

Once gathered from the CF Trust discussion forum, user posts were analysed using framework thematic analysis (Ritchie & Lewis, 2003). Choice of analysis approach was guided by two factors. Firstly, the primary aim of the study was to explore the topics under discussion on the forum rather than to provide a representation of the experience of forum users or analysis of their conversation per se. Secondly, the nature of data collection meant that all data was cross-sectional and collected prior to analysis and the researcher was unable to ask further questions to explore emergent themes. Thus, thematic analysis was felt more appropriate than a phenomenological or grounded theory methodology. More specifically, framework analysis was selected in order to provide a structured and transparent method to approach data analysis (Ritchie & Lewis, 2003; Smith & Firth, 2011). Although framework

analysis has been used in a relatively 'top-down' manner within applied policy research (e.g. Srivastava & Thomson, 2009), the original developers highlight that its use was intended to be dynamic and flexible (Ritchie & Lewis, 2003). The utility of framework analysis within healthcare research has become increasingly recognised in recent years (Smith & Firth, 2011). Framework analysis involves moving through a number of stages including familiarisation, identifying a thematic framework, indexing, charting and mapping and interpretation (Ritchie & Lewis, 2003). Although these stages can be sequential, researchers benefit from moving forwards and backwards through each data stage in order to ensure that analysis remains close to the original data and misinterpretations are minimised (Ritchie & Lewis, 2003; Smith & Firth, 2011). Each phase of analysis will now be described.

5.5.1.1 Data management and familiarisation

All posts made by each consenting forum user were collected and arranged in chronological order. A subset of six participants' posts were selected which were felt to present a varied range of discussion topics and posts. The researcher became familiar with the content of the posts by reading and re-reading.

5.5.1.2 Identifying a thematic framework and indexing

During the latter stages of familiarisation, the researcher began to note recurring topics of discussion present in the data. A further reading of the subset of posts was carried out with the specific purpose of building a list of the topics covered and the characteristics noticed within them (e.g. whether posts were seeking information, providing advice, sharing experiences etc.). Once it was felt that the data was adequately represented by the identified categories, the researcher wrote each of these on a separate piece of paper. This allowed categories to be moved freely and for different organisational groups to be explored. With frequent referencing back to the raw data, this process allowed the development of the initial indexing framework which would be used to code the remaining data which was not

included in the initial subset for familiarisation. An example of how the themes were derived from the original data is presented in Table 5.1. At this stage, it was important for the framework to remain flexible to ensure that when approaching the whole data set, categories not fitting well into the initial framework could be logged and revisions made. A copy of the initial indexing framework is included in Appendix G.

The indexing phase involved applying the thematic framework to the whole data set. Each sentence, phrase or paragraph of each post was considered in turn and coded in accordance with the indexing framework. Throughout this process, the indexing framework was revised in order to capture new topics not previously identified within the familiarisation subset of posts. A copy of the final indexing framework can be found in Appendix H.

5.5.1.3 Charting and mapping

Thematic charts, also known as matrices, were created to enable a view of the whole data set using computerised spreadsheet software. Seven charts, one for each main theme were developed to include key words, quotations and summarised content for each participant within each theme sub-topic. Each index category was represented by a column and each participant represented by a separate row. This presentation method allowed for comparison across cases (i.e. forum users) to ensure consistency within and distinction between themes. Reviewing data in this way also brought attention to topics which could be merged together into a broader category whilst remaining true to the raw data.

5.5.1.4 Interpretation

The final stage of framework analysis was to develop a way of summarising and interpreting data in terms of identified patterns, concepts and explanations. The matrices of data enabled the researcher to gain an overview of themes, tone of posts and individual user narratives.

Table 5.1 Example process for arriving at a theme

Participant	Post extract	Preliminary thoughts	Category	Initial theme
Female 25 years old	*'I usually have a well behaved port and have got used to its funny little ways and how it usually behaves.'*	Getting used to changes in treatment.	Adjustment and transitions	Psychosocial processes and issues
	'Same goes for explaining to friends why I need IVs when I outwardly don't look that ill. I love my friends dearly, but there's lots they don't understand.'	Explaining to others but them not fully understanding.	Friends not understanding	
	'I usually hide [my cough] quite well though, I have breathing techniques I use to take it away unless I'm really bad. Then when I do have a normal coughing fit in public, everyone starts to panic and fuss over me, not accepting the "this is normal, please leave me alone, I'll live". I hate it lol'	Actively trying to hide coughing symptoms.	Hiding symptoms of CF	
		People noticing symptoms which are not normal to them.	Feeling different	

Key products of this phase included the creation of a data display diagram to visually represent the key themes and their inter-relations. Displaying data in this manner aids researchers in understanding, analysing and communicating to others the themes and patterns

which have emerged throughout the analysis process (Miles & Huberman, 1994; Ritchie & Lewis, 2003). In addition to this visual representation of data, a written explanation of themes and results of the analysis was also developed.

5.5.1.5 Software

Qualitative analysis was aided by using Nvivo 8, a computer programme designed by QSR International (2008) to help organise and track the analysis process. Once imported into Nvivo, codes were assigned to text in line with the indexing framework developed during the earlier stages. Computerised spreadsheet software was used to create tables during the charting process to illustrate themes and case-examples from the qualitative data.

5.5.1.6 Ensuring validity and quality in qualitative research

In order to maintain validity and rigour within the qualitative analysis process, a number of strategies were used. The researcher aimed to carry out analysis in a transparent and reflexive manner to ensure that the processes followed were clear and considered (Lewis & Ritchie, 2003; Yardley, 2008). Although it was not possible to obtain a full second opinion on applying codes to data, two researchers were consulted throughout the analysis process. Discussion was carried out around the data, emerging themes, potential refinements, development of data displays and ensuring consistency and coherence throughout the analysis (Yardley, 2008). A reflective approach was taken during all phases of research in order to remain open to new themes as well as potential researcher influences and contextual factors which may have impacted on the analysis process (Lewis & Ritchie, 2003; Yardley, 2008). Finally, whilst creating the data display and exploring inter-relations between themes, the researcher made efforts to identify cases within the data which provided contradictory examples to the developing understanding.

6 STUDY 2 RESULTS

Six key themes were identified from the analysis process to encompass the index topics present within the forum post data: regular or daily CF treatment; hospital experiences and procedures; changes in health and progression of illness; daily living and occupation; psychosocial processes and issues; and the future. A seventh set of index categories were also grouped together which identified the tone and nature of the post, for example, whether the author was seeking information, advice and sharing experiences or giving empathy, encouragement or reassurance. Figure 6.1 visually represents the main thematic groups and their sub-topics. It also includes reference to the general tone of posts across themes. In the following sections, each key theme, the tone of forum posts and comment on the individual users' narratives running through the posts will be described.

6.1 REGULAR OR DAILY CF TREATMENT

The forum posts revealed regular discussion about aspects of routine CF treatment and its considerable demands on patients' time and effort.

> P1, 25 year old female:
>
> > *Just staying well and maintaining my health takes hours of treatment a day, a lot of energy and focus and the help from everyone around me.*

Posts in this theme covered topics of oral and intravenous antibiotics, nebulisers, physiotherapy, diet and digestive enzymes. Some users sought clarification on dosage, timings of treatment, how medications worked or the different types of treatments available. Other users shared their own experiences, provided information or suggested advice. A number of threads introduced the pros and cons of particular types of treatment, for example providing explanation of different physiotherapy devices or sharing experiences of different

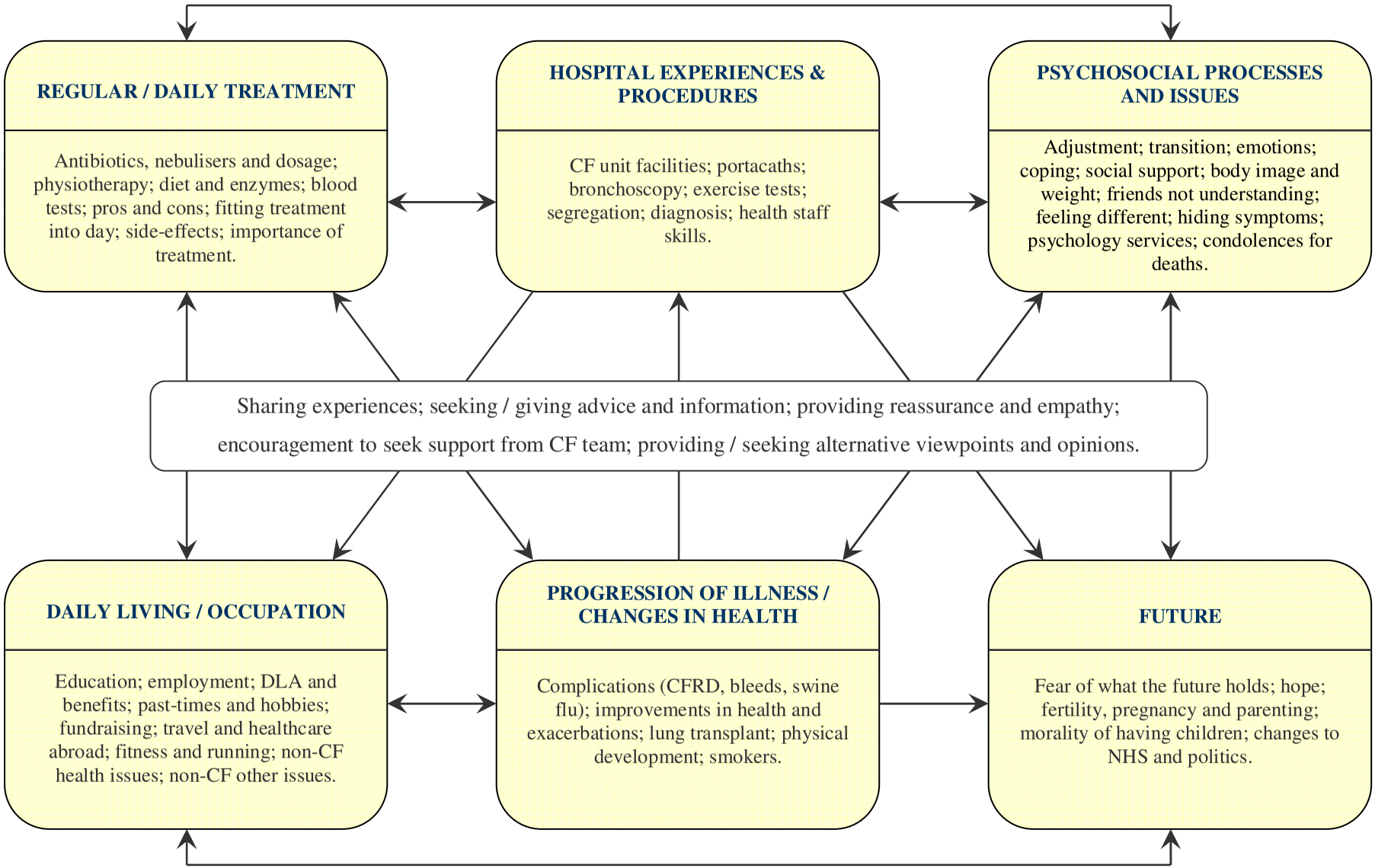

Figure 6.1 Main themes resulting from framework analysis of discussion forum posts

concentrations of hypertonic-saline (a solution inhaled to help mobilise mucus prior to physiotherapy). Included in the 'cons' of treatments was the discussion of side-effects from some medications, such as steroids or specific antibiotics.

Four participants made reference to the difficulties of fitting treatments into everyday life, with recognition that treatments were occasionally missed.

P1, 25 year old female:

> *I always get up 2 hours before I leave the house so I have time to neb, physio, shower, get dressed and eat. Sometimes I don't fit in the eating bit so grab something as I'm leaving or pay a quick visit to a shop. This morning though had to leave at 6.30 and woke at 5.45 so I've missed physio and neb and food.*

P11, 36 year old female:

> *I totally understand where you['re] coming from. I have to get up at 6am just so I can drop my son and his m8s in to school.*

Amongst discussions about CF treatment there was an emphasis on the importance of adhering to particular treatments or associated monitoring procedures. Some posts used empathy, shared experiences and offered advice in a gentle manner, whilst others appeared more blunt in their approach. Multiple posts focussed on the use of one particular antibiotic, Tobramycin ('Tobi' or 'Tobra'). If blood levels of this drug are not carefully monitored, serious and permanent side-effects, such as deafness, can occur.

P9, 27 year old female:

I'm also not surprised that they won't give you tobra if you wont let them take bloods. It would have been highly irresponsible for them to do so.

P8, 27 year old female:

Giving blood is not a big deal, yes it's a pain, and it's even more annoying when they miss, hit and pull out because of nerves, take the wrong vials, lose it or do the wrong test. But it's still necessary. I'm assuming you have a port if you hate needles this much?

Other post emphasised the importance of adherence to dietary recommendations after lung transplantation to minimise the risk of becoming ill.

P13, 48 year old male:

Any normal person will get a mild case of the runs. A person on immunosuppressants will get a very bad case of the runs, and in a couple of hours the bacteria will rip through their system and kill them.

In contrast to the posts highlighting the more serious notes about CF treatment, some forum users chose a lighter, more humorous way to comment on treatment. One example of this provided a parody description of a physiotherapy device known as the 'vest' which vibrates to help mobilise mucus in the lungs during chest physiotherapy.

P5, 32 year old male:

Want to improve your physio? Maybe you've even thought about buying the VEST? Don't! [P5] has a better, cheaper and ultimately more

satisfying idea. Instead of buying 'the vest', a very costly device, why not selotape a kitten to the front and back of your chest? Get the kittens to purr and you have your own cheap vest.

This post was received well by other forum users and potentially acted to provide some comic-relief from discussion around the more serious topics of treatment adherence and side-effects.

6.2 HOSPITAL EXPERIENCES AND PROCEDURES

Topics falling within this second theme centred around two areas: a) the experiences of inpatient admissions and hospital clinic visits and b) medical procedures which are not part of routine daily CF treatment.

6.2.1 Hospital experiences

Forum users discussed different CF units, their available facilities (for example, internet or mobile phone signal), the way clinics are run, segregation and infection-control policies. Posts outlined the resources available at and characteristics of particular units and offered reassurance to people seeking support prior to admission.

P9, 27 year old female:

Sorry to hear you have to go in, but you'll be glad to hear the new ward is really very very good. The staff are friendly and helpful. The rooms are ensuite with Digital TV and free internet… The ward has it's own kitchen and chefs… There is an exercise room with a TV… Take the chance to have a little rest and be waited upon!

Infection control strategies at different CF units were discussed alongside sharing views on segregation policies and their impact on cross-infection. Participants seemed to acknowledge that segregation was part and parcel of CF treatment and generally spoke positively about its contribution to reducing the likelihood of cross-infection and development of drug-resistant strains of infections. However, the difficulties with segregation were acknowledged, particularly for families where more than one child has CF or where patients with specific strains of infection were uncertain about being allowed access to new CF unit facilities. Personal viewpoints about segregation were voiced; some users suggested that everyone with CF had a duty to the wider CF community to prevent cross-infection and others indicated that their decisions to limit contact with other CF suffers were for their own health.

P13, 48 year old male:

> *If its just a very few couples and people from the same family infecting each other then that is bad for them, but not too bad for the CF community as a whole. But if I met a person with Cepacia, then met you, then we both went on to meet other pwcf (*people with CF*), then lots of people would end up with Cepacia. And if we are all being careless and meeting up, then that is bad for everybody.*

P17, 24 year old male:

> *Ultimately we all have to make our own decisions. My decision is to keep away from you all – for my sake and yours :D*

6.2.2 Medical procedures

The data set indicated a frequent exchange of knowledge and experiences between CF patients regarding medical procedures such as having a portacath ('port') fitted, having a

bronchoscopy or the addition of new tests to CF clinic visits. Advice was also provided to parents of children with suspected CF regarding the different diagnostic procedures, their limitations and range of outcomes. Some participants were seen to seek support and reassurance for their anxiety about upcoming procedures and to gain insight into the experiences of others who had similar procedures. Fellow forum users provided this, along with tips for coping with procedures, descriptions of personal experiences and, on one occasion, photographs of a port to help allay others' fears.

P7, 28 year old female:

I decided to have my port fitted last September and actually had it done 4 weeks ago. It looks ok and doesn't show as much as I thought it would as I am skinny… I am more than happy to email you some pics…

Skills of health staff were mentioned in relation to general CF care and specific medical procedures. Experiences of health staff skills were discussed in the context of inpatient stays and receiving routine blood tests. Participants expressed frustration at repeated failed attempts at inserting cannulas or taking blood, at times when procedures needed to be repeated and at the lack of CF-specific knowledge and experience of some staff.

P1, 25 year old female:

I have one vein that always bleeds, but it's right next to a knuckle and it really really hurts, but not everyone will use that vein as they prefer to attempt the non-existent veins in my arms 50 times first, which hurts a lot more.

Forum users shared their own experiences and provided support for others in relation to bad experiences. Some users acknowledged that mistakes are sometimes unavoidable and that

repeated tests or procedures were frequent in CF care. Users identified with others' experiences through sharing similar stories of frustration or the use of humour.

P5, 32 year old male:

Getting jabbed a billion times is all part of the territory with CF… tad annoying...

P8, 26 year old female:

I was sedated by an inexperienced doctor and merry hell broke loose with my diabetes, I was LOW and boy was I craaaanky.

6.3 CHANGES IN HEALTH AND PROGRESSION OF ILLNESS

In addition to discussing CF treatment and associated medical procedures, there were many topics noted which addressed changes in health over time. Forum posts mentioning such changes (for better or worse), physical development and growth and lung transplant were encompassed by this theme.

6.3.1 Changes in health

Participants discussed and shared experiences of fluctuations in health and complications such as CF related diabetes (CFRD), respiratory infections, bleeds in the lungs and swine flu. Users who reported their health to be at a lower level often mentioned being admitted more frequently to hospital and receiving more treatment. Those who were experiencing health difficulties sought advice and alternative opinions on potential causes of exacerbations, avenues for treatment and support in coping with worsening symptoms. Information was provided by some participants on common complications which may develop over time in people with CF, for example CFRD, or the contraction of aggressive and hard-to-treat

infections such as pseudomonas or aspergillus. A particular user shared his extensive experiences of embolisation ('embo') procedures, which are used to treat bleeds into the lungs, and the continual battle for maintaining health.

P5, 32 year old male:

My bleeding has really slowed due to embos, went for a good couple of months without a bleed recently… It can be annoying bleeding, but recently I've found it especially annoying because it creates an environment for infection and therefore you feel you are constantly battling a light infection and trying to stave off IVs.

In terms of short-term aggravation of symptoms, some participants mentioned the topic of smoking. Users expressed anger towards members of the public who ignored 'no smoking' signs whilst others warned fellow forum users of the health consequences of smoking when diagnosed with CF. A minority of posts mentioned their current good health despite some decline since childhood, reporting that they felt lucky to be doing well.

6.3.2 Physical development and growth

Physical development and growth topics were included in a number of posts. Advice was sought about typical growth patterns, i.e. if later growth spurts occurred for people with CF and expressing concerns about being shorter in stature than peers. Other users sought advice or shared their concerns about menstruation and the affects that hormones had on the ability to cope with CF treatment.

P12, 23 year old female:

I was still slightly concerned about my height and seemed to pretty much hit my final height… age 12 and am 4ft 10… my paediatric CF dr

predicted my height etc based on other members of the family and patterns in my own growth etc. My Gran was 4ft 11, mum is 5ft 2 so really my height isn't bad for my family…

6.3.3 Lung transplant

Lung transplant was a topic which arose in relation to several main themes – CF treatment, medical procedures and the future. It was felt that, in light of lung transplant ordinarily being suggested at a point of particularly poor health and the fact that it can lead to dramatic changes in respiratory function, this topic was best represented alongside other topics related to changing health and disease progression. Discussions around transplant varied in terms of the position of the participant. Some were coming to terms with the suggestion of lung transplant by their team or seeking information on the transplant process.

P15, 20 year old female:

I think it's also the fact I spent half of last year on IV's including quite a few admissions to keep me at that lung function which has made them suggest transplant. In my head I think I'm too well at the moment anyway, it's just the fact I know it will have to happen in the not too distant future.

Other users shared experiences of being post-transplant, provided information on aspects of the transplant process and discussed practical changes or dilemmas they currently faced, for example on diet restrictions, implications for employment or benefit reassessment.

P16, 28 year old female, post-transplant:

I have decided that it's time for me to look for a job in my degree [subject area]… My question is should I mention the whole transplant thing? Would some people see it as a negative thing when hiring? I feel like I need to mention it because it explains why I'm in an office job…

Ok things I've been told I can't eat that pop into my head right now – sushi, smoked salmon, rare meats, pate, tiramisu (sp?), some cheeses, natural yoghurt, soft ice cream, grapefruit.

The presence of several people on the forum who had undergone successful lung transplant led to multiple posts of congratulation and well-wishing. However, not all forum members were so fortunate and members posted their memories and condolences for those who had lost their battle at some point on the transplant journey. One forum user shared a conversation he had with a fellow CF sufferer who was awaiting transplant and who later died:

P5, 32 year old male:

I was speaking to him last week about transplant and how he felt about it. I asked him, as it was one of my greatest concerns about transplant, whether he feared dying. He said he didn't fear it, he was more worried about the people he would leave behind. It's nice knowing he wasn't scared or worried about what could happen.

6.4 DAILY LIVING AND OCCUPATION

Forum users frequently had discussions around issues of everyday living and occupation. Most topics within this cluster were discussed in the context of CF with specific references to education, employment, Disability Living Allowance (DLA) and benefits, travel or living abroad and fitness and running. Other topics within this group which did not include direct discussion about the impact of CF included fundraising, past-times and hobbies, non-CF health topics and miscellaneous questions not related to CF.

6.4.1 Education and employment

Participants discussed their varied experiences of school and university, special arrangements that were provided for them during exams and how they felt their CF impacted on their education. Some users expressed their ambivalence about accepting special exam conditions as they wanted to be treated the same as their peers but were conscious that coughing during exams could disturb other students. A number of participants exchanged memories from school and university, some positive, some negative and some with a twist of humour:

> P5, 32 year old male:
>
> *I regularly had coughing fits during lectures and the lecturer would stop the lecture to see if I was alive. I often couldn't speak, so just gave the thumbs up, it became a running joke… in a good way.*

Advice was provided to parents in supporting their child with CF to make realistic decisions about their education and career path. Patients with CF shared their employment status, reasons for choosing their respective careers and the influence their health played in career decisions. Additional common topics pertinent to those seeking work or considering career options were: the disclosure of CF diagnosis to potential employers and explaining gaps in

employment due to ill health. Forum users contributed their personal opinions on when they felt was the most appropriate time to disclose diagnosis and offered advice to those who were currently making decisions about this. Participants also discussed whether it would be detrimental to gaining employment if they were to disclose true reasons for gaps in their CV. Some users advocated being upfront about this, whilst others felt this would decrease the likelihood of being offered employment and that an alternative reason for gaps, if available, should be given instead.

P17, 24 year old male:

I never mention CF at interview – I agree with [another forum user] that it's expecting too much of an average manager to figure out the implications. I've never actually been asked any health questions at interview anyway so its not like I've felt that I was evading the question.

6.4.2 Disability Living Allowance (DLA) and benefits

Participants contributed to a number of discussions about benefits and DLA. Some posts sought information about the types of benefits people with CF may be entitled to and the best ways to explain the nature of CF when filling in application forms. Complementing these posts were others which shared experiences of receiving benefits in the context of personal circumstances and clarified eligibility for the different types of benefits. Furthermore, comments were posted which explained to others that benefits and carer allowances made it possible to live independently from parents despite considerable health needs. An area of concern for people with CF raised within posts about DLA was the government changes in DLA assessment. Links were provided to external websites which outlined the move to 'capability assessment' and these were followed by discussion of worries that the new assessment would not adequately account for CF and its impact on patients.

6.4.3 Travel and healthcare abroad

Considerable discussion was noted around the topic of travel abroad. Participants sought advice on travel insurance and the logistics of travelling with CF, such as how to keep medications cool during transit or arranging oxygen to be accessed at destinations abroad if required. Forum users offered their own experiences and provided advice on these topics. Some posts indicated the importance of being honest when getting travel insurance to ensure that potential health needs are fully covered if something were to go wrong whilst away from home.

> P13, 48 year old male:
>
> *I think the thing to declare is 'post transplant (as the result of cystic fibrosis)'… If you don't inform the insurers then the premiums you paid will be worthless and you could end up with the sort of expenses that bankrupt people.*

In addition to advice and information about holidays, discussions were also centred on living abroad and the CF healthcare available in other countries such as France, Denmark, Sweden and Australia. Participants shared links to appropriate websites and information they had found when considering a move abroad themselves.

6.4.4 Other topics

In addition to the key issues outlined above, data falling within this theme covered topics of fundraising, fitness and running, hobbies, non-CF health and non-CF general issues. These posts included discussion about how to organise and advertise fundraising events and sharing training advice to prepare for running events, such as half and full marathons. There were also more general discussions around shared hobbies or preferred pastimes such as going to the theatre or computer games. Some participants sought information and advice on non-CF

health issues including migraines, HIV and dental advice. Other unrelated general issues were also discussed, such as home heating systems and ideas for birthday celebrations.

6.5 PSYCHOSOCIAL PROCESSES AND ISSUES

The fifth main theme which emerged from the forum post data concerned psychosocial issues. Forum post contents fell into several sub-topic domains consisting of: adjustment and transition, interpersonal relationships, emotions, coping, body image and weight and social support.

6.5.1 Adjustment and transitions

Participants made reference to adjustments and transitions with regard to a range of topics. Some posts focused on practical adjustments such as getting used to new treatment methods or procedures, for example having a portacath or using oxygen at home. Other participants shared their experiences of adjusting to changes in health: either coming to terms with the suggestion of having a lung transplant or adjusting to life post-transplant. Transitions tended to be mentioned in the context of moving from paediatric to adult services or when discussing transition of adolescents to take more responsibility for their own treatment. Participants provided empathy and reassurance, shared personal experiences of transitions and gave support to other forum users facing similar stages in life.

P13, 48 year old male:

> *Of course you're scared, and there's nothing selfish about it! You know the paediatric team, you know how good they are, and you know you can trust them! You can't say the same about the adult team. You don't know them… It sounds like your son is beginning to take control and responsibility for his treatment. This is a very encouraging sign.*

6.5.2 Interpersonal relationships

Posts falling into this category introduced some interesting insights into the complexities of friendships for people with CF. Participants reflected on feeling different to their peers and having a hidden illness. One of the giveaway signs of CF for which concern was expressed was in coughing at school, university, work or in public. A number of forum users felt that the separate arrangements for exams whilst at school and university did not act to single-out people with CF because students with other difficulties, e.g. dyslexia, also required such additional support. Nevertheless, other participants expressed their concerns about being seen as different. Forum users explained their efforts to suppress coughing and hide visible signs of having CF, such as covering up a protruding stomach, portacath or cannula. Not only did this seem to be important for fitting in with peers, but also in hiding symptoms from family members or the CF team. Interestingly, two participants made reference to wanting to hide reminders of CF from themselves.

P3, 25 year old female

> *I think it's undisputed that having [a port] makes IVs easier which sounds great. But like you my big concern is having it there, seeing it every day even if I'm well it would always be reminding me.*

P15, 20 year old female:

> *I am so guilty of the "I'm fine!" response when anybody asks me how I'm doing. Even my CF team raise their eyebrows and tilt their heads when I say "I'm fine"… knowing that really means I'm terrible and need IV's asap, they know me too well now!*

Despite participants' attempts and desire to hide their symptoms and the reality of CF from friends and family, threads on the forum indicated that they felt frustrated when it became apparent that friends did not understand what it meant or felt like to have CF. One participant shared her experience of telling her friends that she had been put on the lung transplant list:

P15, 20 year old female:

Recently transplant has been suggested by my clinic and I've been a bit emotional about it. Wanting them to understand so I had somebody to talk to about it, I got a couple of my close friends together and broke the news. During their reaction one of them, looking puzzled, said, "oh, well I kind of thought you were always on the transplant list anyway?!" to which the others nodded and agreed.

Similarly, an older participant who had children shared her experience of other mothers when arriving at the school gates.

P11, 36 year old female:

…they smile and shake their ignorant heads. While I'm rushing around in the morning like a coughing, spluttering banshee they are sipping their coffee having a fag.

6.5.3 Emotions, coping and psychology services

Many emotions were expressed across the forum posts in a number of contexts. As would be naturally expected, anxiety and fear were commonly reported when discussing decisions about trying new treatments, upcoming procedures or when adjusting to changes in health. Low mood was mentioned by two participants who described identifying with other CF

sufferers in magazine articles or films which acted as a reminder of the potential trajectory of CF in the future. Anger was mentioned in a number of posts in relation to smokers, especially the fact that they were voluntarily damaging their lungs, that they may receive lung transplants instead of someone with CF or that smokers' lungs may be used for transplant into someone with CF.

In relation to coping with the realities of having CF, the demanding treatment regime and accompanying emotions, posts pointed readers in the direction of available psychology or self-help services and also suggested possible ways of coping. Some posts focussed on providing practical advice such as becoming as educated as possible about CF, seeking support from the CF health team and making treatment part of a habitual, daily routine. Strategies for coping with emotional aspects of CF were also offered, for example, remaining realistic in expectations, accepting personal limitations, trying to stay positive and hoping for advances in medical science. Some users shared their own experiences of having difficulties coping in the past and the detrimental effects of focussing on the worst case scenario.

P13, 48 year old male:

> *When I was 17, I felt very similar… I assumed there was no reason to make plans or get married, and that having kids was the most dumb thing I could think of. It turns out that assuming I was going to die was the mistake – the safe mistake, the sensible mistake, but still a BIG mistake.*

6.5.4 Body image and weight

Although attempts were made to hide visible indicators of CF, one common CF characteristic discussed on the forum was not possible to hide: being small in size compared

to peers. Although some participants offered reassurance that growth spurts tended to occur later on for people with CF, some forum users found their appearance frustrating. Participants were able to vent their annoyances through sharing experiences of being asked for ID when going to see films, buying alcohol or being teased by work colleagues.

P12, 23 year old female:

The biggest guy at work wanted to put me on the top shelf in a cupboard just 'cause he could.

In contrast to the annoyances expressed on the forum about fluctuating weight and small size which were mentioned in a generally light-hearted way, some posts sought support from others with weight difficulties which had become health problems in themselves. Other forum users offered support and advice to those struggling to put on weight and those who were fearful of gaining weight.

6.5.5 Social support

Across many posts and themes, it was noted that empathy, reassurance and encouragement were frequently used to support fellow users at times of difficulty. Many posts centred on wishing people well, sending congratulations or wishing others a happy birthday. However, there were also numerous messages of sorrow, support and condolences for the family and friends of fellow forum users who had died. Despite the life-limiting nature of CF and the recognition of this by patients, some posts expressed the shock of losing a friend.

P17, 24 year old male:

Such a shock. Wasn't expecting this at all. My thoughts go out to his family and many friends.

Many of the posts for those who have lost their lives to CF end with a touching phrase which seems to have been adopted by the forum to wish people well in death:

P12, 23 year old female:

Really sad to hear this. My thoughts go out to his family and friends, breathe easy.

6.6 FUTURE

Throughout the analysis process, themes emerged which seemed to make reference to the future. The index categories were felt best described in terms of fear and hope, pregnancy, fertility and parenting with CF, and NHS and political changes.

6.6.1 Fear and hope

A number of discussions expressed a fear of what the future may hold, particularly in terms of declining health and its consequences. Some feared the risks involved with having a transplant whereas others worried about the impact on daily living or their ability to cope with the process.

P15, 20 year old female:

I've not been able to sleep or function properly since Tues, just cannot think of anything but transplant and what the future holds now.

In contrast to the posts expressing fear, there were also those which presented a more hopeful view of the future. These posts encompassed small specific hopes, e.g. for a portacath to continue working well, and larger hopes such as those for a long life in light of current good health or for medical advances to find a cure for CF. Some offered hope

through posting inspirational quotes whilst one forum member reassured others by using increasing life-expectancy statistics.

P13, 48 year old male:

It looks likely that even without breakthrough treatments, most kids born these days will live to see their 50th birthdays.

6.6.2 Fertility, pregnancy and parenting with CF

Several threads contained the sub-theme about future plans to have a family. Within these posts, participants mentioned fertility, the health consequences of pregnancy and coping with parenting with CF. Forum users sought information and advice on many aspects of becoming a mother and also contributed views about the difficulties which might be faced in the role of being a parent with CF. Members discussed their fears that future declines in health may mean that pregnancy was too risky.

P12, 23 year old female:

If I were to start on a downward spiral, I would have to reconsider having a family, but what also worries me is that I am well enough to have a family but that the stress of pregnancy and various other factors after birth etc could cause me to decline.

Furthermore, a particularly emotive topic in relation to being a parent was raised; the morality of having children when the parent has CF. This brought numerous viewpoints and responses as people considered the potential impacts that CF may have on a person's ability to cope with parenting roles. Some people reflected on their own childhood experiences of losing a parent to an illness and felt that doing this to a child was unfair. Others felt similarly

that being a parent with CF would prevent meeting children's needs and lead to unbearable levels of guilt. Despite concerns, one participant felt that, providing her health was good enough, she would opt to have children but recognised that listening to the CF health team would ultimately guide her decisions.

P3, 25 year old female:

Is it really fair to knowingly bring a child into a world where they have to amuse themselves whilst you do treatments, where you are in and out of hospital, where they will probably lose a parent early in life??

P12, 23 year old female:

If I am still well enough when the time is right then I will try and have a family. If I'm not… then I won't, but it will be extremely hard on me emotionally.

6.6.3 Future changes in the NHS and DLA

The final sub-topic referencing the future was the discussion of governmental decisions which would impact on the provision of NHS health care and DLA benefits. Posts raised awareness of issues and provided web-links to further information about proposed and planned changes. A forum user raised concerns about changes to DLA and the impact these may have on people with CF.

6.7 TONE OF POSTS AND PARTICIPANT NARRATIVES

As mentioned within the section on social support, it was noted throughout analysis that posts generally had a supportive tone which provided empathy, reassurance and encouragement to other forum users. Those posts seeking support or information sometimes

covered difficult and emotive issues. The willingness of users to share personal experiences indicates a sense of safety within this online community. Numerous posts expressed thanks to other forum members for the provision of support, information and advice.

Additional observations were made concerning the narratives of each forum user included in the study. It was evident through examination of the thematic matrices that some users' posts reflected concerns or questions regarding particular issues which were pertinent for them at the time, such as lung transplant or declining health. On the other hand, a number of users tended to be the providers of advice, support and information to those seeking it. They shared their own experiences, educated others and provided web-links to external sources of further information. Some forum members appeared to be known amongst the community for having a wide knowledge base on particular subjects, and were called on for such information when needed. Additionally, it was noted that some members' roles involved providing comic relief and to instigate light-hearted interactions. This perhaps acted to lift the general mood on the forum and provide an additional coping strategy for the realities of living with CF.

6.8 SUMMARY

Framework analysis of the forum posts revealed a diverse range of topics. Six main themes emerged from the indexed data which covered the daily and routine aspects of CF treatment, hospital experiences and medical procedures, daily living issues including employment and education, changes in health and disease progression, psychosocial processes and issues and coping with the future. The data illustrated the supportive nature of the posts and provided insight into the people who make up this community. Individuals shared experiences and offered advice, empathy, encouragement and reassurance. They provided information and support both for CF-related issues and those with no relation to their condition. Forum users were noted to use the resource differently over the period for which data was collected.

Some were seen to be seeking specific support in light of particular life-events or health changes, whereas others more consistently provided information, advice and shared their own experiences. The forum creates an opportunity for people with CF to interact without the risk of cross-infection. Participants are able to meet and discuss issues in a large community where others have first hand experience of what they are coping with. Due to segregation policies, this would not be possible face-to-face.

7 DISCUSSION

The first study aimed to answer two primary research questions. These were to determine the levels of mental health symptoms, HRQoL and self-esteem in adults with cystic fibrosis who made use of an online discussion forum and whether any significant correlation existed between these variables. To the author's knowledge, no previous studies have explored self-esteem and its relationship with mental health and quality of life in adults with CF. The second study aimed to provide a qualitative insight into the topics arising on an online discussion forum for people whose lives are affected by CF. Although some studies have explored the use of specifically designed online support groups (Griffiths *et al.*, 2009), there appears to be no representation of CF within this literature. The following sections provide discussion of the results from both studies within the context of current research literature. The clinical implications and limitations of the findings will also be addressed, along with consideration of directions for future research.

7.1 MENTAL HEALTH, QUALITY OF LIFE AND SELF-ESTEEM

Thirty per cent of the current sample fell above the clinical threshold of mental health symptoms when assessed using the CORE-OM. This percentage is higher than would be expected in the general population (Singleton *et al.*, 2001) and is line with other published studies which conclude that adults with CF present with elevated levels of mental health difficulty (Cruz *et al.*, 2009; Goldbeck *et al.*, 2010; Havermans *et al.*, 2008; Modi *et al.*, 2011; Riekert *et al.*, 2007). However, the finding is in contrast to the literature which provides evidence against elevated mental health difficulties in adults with CF compared to those without (Anderson *et al.*, 2001; Pfeffer *et al.*, 2003; Szyndler *et al.*, 2005). In terms of self-esteem, the total mean score of 29.89 was comparable to scores presented in research by Abbott *et al.* (2000, 2007) who explained that in general, self-esteem for the CF groups was comparable to healthy control groups.

Although levels of mental health difficulty and self-esteem were described at similar levels to previous research, this was not the case for quality of life scores. There was notable variation between average sub-domain scores on the CFQ-R which ranged from 46.96 for physical functioning to 65.61 for role functioning. Scores for all four sub-domains were considerably lower than those reported in other studies (Casier *et al.*, 2011; Sawicki *et al.*, 2011; Szyndler *et al.*, 2005). There are a number of possible explanations for the lower levels of HRQoL in this study, one of which may be found in the differences between sample population characteristics compared to other studies. For example, both Casier *et al.*'s (2011) and Szyndler *et al.*'s (2005) sample involved much younger participants (mean age 18.4 and 15.0 years old respectively) who had better lung function (mean FEV_1% predicted scores of 83.1 per cent and 72.4 per cent respectively). Although age has not been shown to influence quality of life and psychological well-being significantly (Havermans *et al.*, 2008; Riekert *et al.*, 2007), those with more severe disease are more likely to experience psychosocial difficulty and lower quality of life (Gee *et al.*, 2003; Oxley & Webb, 2005; Pfeffer *et al.*, 2003). Participants included in the current study had a mean FEV_1% predicted score of 61.7 per cent, in the upper end of the moderate disease category. When looking at the proportion of participants falling within each category, the majority are within the moderate or severe category of disease (62 per cent). From this, it may be considered that the lower HRQoL scores across all domains may be partially due to the severity of disease in the current sample. However, this explanation does not hold true when comparing to the study by Sawicki *et al.* (2011). Their large sample of 631 adults with CF were of a similar mean age (26.9 years, SD 9.7) and severity (FEV1% 61.0, SD 23.0), yet mean scores for the four CFQ-R domain scores were approximately 20 points higher: physical 67.9 (SD 27.4), role 81.1 (SD 19.4), emotional 76.3 (SD 20.0) and social 71.6 (SD 17.9). In this case, an alternative contributory factor to the difference may be grounded in the unequal gender split and overrepresentation of females within the current sample.

Prior research has established that females with CF tend to fair worse medically than males (Nick *et al.*, 2010; Stephenson *et al.*, 2011). Furthermore, psychological research has established a number of gender differences, for example, females have been shown to experience higher levels of anxiety and depression (Anderson, *et al.*, 2001; Goldbeck *et al.*, 2010; Modi *et al.*, 2011), poorer HRQoL (Arrington-Sanders *et al.*, 2006; Gee *et al.*, 2003) and lower self-esteem (Abbott *et al.*, 2000; 2007). The current study supports these findings and presents data indicating that females experience considerably more difficulties in a number of areas of psychological functioning and HRQoL. The exploratory analyses conducted within Study 1 show that females scored significantly lower on measures of emotional functioning, social functioning and self-esteem and higher on presence of mental health symptoms. Recent papers have highlighted the complexity in the interrelationships between mental health, HRQoL and social functioning in males and females (Abbott, 2009; Brucefors *et al.*, 2011). It seems that females experience lower levels of HRQoL and have more difficulty managing symptoms, both physical and psychological, than males (Abbott, 2009). Additionally, females who perceive their situation as less manageable have greater difficulty with anxiety, depression and social dysfunction (Brucefors *et al.*, 2011). Gender influences in CF are emerging to be a complex picture and it would be useful for future studies take a longitudinal approach to examine the impacts of physical health and psychosocial variables over time.

It seems that the female dominated sample in the present study may be one possible explanation for the lower mean HRQoL scores compared to previous studies. However, it is also possible that low scores are a reflection of the particular sample recruited. It may be that those with poorer quality of life were particularly attracted to taking part in the study. Alternatively, online resources can provide a convenient and anonymous environment which is available for those with access to the internet regardless of health status and mobility. As

such, it may be that forums are frequented more often by those who have more physical health restrictions or who are less able to engage with aspects of 'real-life' and therefore have poorer HRQoL. This would contribute to the lower levels of HRQoL reported within the current sample.

7.2 CORRELATIONS BETWEEN VARIABLES

Correlation analyses between outcome variables revealed that higher self-esteem was related to better mental health and that better HRQoL in all four domains. These findings are somewhat intuitive and the significant relationships between higher mental health difficulty and lower levels of quality of life in all domains, including social functioning, are consistent with the results of previous research in adolescents and adults with CF (Casier *et al.*, 2011; Havermans *et al.*, 2008; Quittner *et al.*, 2008; Riekert *et al.*, 2007; Szyndler *et al.*, 2005).

Less severe CF disease, as measured by higher FEV_1% predicted scores, was associated with better physical, role and social functioning domains of quality of life. However, no significant relationship was found between FEV_1% predicted and emotional functioning, mental health difficulty or self-esteem. FEV_1% predicted is a clinical measure of lung functioning and, therefore, lower scores indicate more severe lung disease. It follows that those people with poorer lung functioning will have more limitations on their physical health, be less able to carry out some roles within families, relationships or employment and be less able to socialise with friends.

There are few studies which have specifically considered the role of self-esteem in relation to mental health and HRQoL in the CF population. Two studies published by Abbott and colleagues (Abbott *et al.*, 2000, 2007) considered self-esteem alongside body image and eating behaviours in adults with CF. Although prevalence of eating disorders was low, their studies indicated a relationship between poorer self-esteem, enteral tube feeds and lower

levels of body satisfaction. The current study indicates links between high levels of self-esteem and better mental health and social functioning in adults with CF. This corresponds well with research from the general population highlighting the protective influence of self-esteem on mental health and, when combined with supportive social circumstances, its positive influence on low mood and coping with serious physical illness (Mann *et al.*, 2004).

Of course, one of the difficulties in using correlation statistics is that causality cannot be inferred. Although there are clearly relationships between the variables of mental health, HRQoL and self-esteem, the influential impacts between them are still unclear and the temporal relationships cannot be deduced in this cross-sectional study. It may be that poorer quality of life due to declining physical health leads to increased mental health difficulty and subsequently results in reduced social functioning. Alternatively, it may be that people experiencing emotional difficulties may function less well socially and experience difficulties adhering to treatments which then impacts on disease status, physical health and HRQoL. A third possibility is that additional variables may be playing a key role in the relationship, for example acceptance (Casier *et al.*, 2008, 2011) or coping strategies (Abbott *et al.*, 2008). Casier *et al.*'s (2008, 2011) studies highlighted the role of acceptance in depressive symptoms over time, but found little influence over anxiety. They revealed that higher levels of acceptance of limitations imposed by physical disease are associated with better psychological functioning (Casier *et al.*, 2008). Moreover, the influence of coping was explored by Abbott and colleagues who found that higher levels of optimism and taking a problem solving approach were associated with higher levels of HRQoL (Abbott *et al.*, 2008). Conversely, higher levels of distraction, or behavioural avoidance, were associated with lower levels of HRQoL (Abbott *et al.*, 2008).

7.3 USE OF ONLINE FORUMS

Exploration of the online discussion forum posts revealed a range of themes spanning a number of domains of life for people with CF. Participants frequently discussed elements of CF treatment, including those which occur on a daily or regular basis, procedures which are less frequent and those related to exacerbations in their condition. Posts were also used to express feelings about particular aspects of life with CF and its associated demands, frustrations and anxieties. Themes acknowledging the impacts of CF on education, employment and the future appeared within the forum posts and many comments reflected processes of adjustment to different stages of life and disease. The topics of conversation present within the analysis have similarities to those presented by three qualitative research projects based on interviews with CF patients which explore the experience of growing up with CF (Jessup & Parkinson, 2010), disclosure of CF diagnosis (Lowton, 2004) and the experience of undergoing lung transplant (Macdonald, 2006).

Jessup and Parkinson's (2010) study invited children and parents to explore their experience of living with CF in unstructured interviews. The authors' phenomenological approach to data analysis revealed eight themes: fright, fear, fight, flight, familiarity, form, future and philosophy and these resonated with those revealed in the present study. Jessup and Parkinson's (2010) participants explained the fright of exacerbations and changes in health, fear of their own mortality and what may happen in the future, including transition to adults services and the inevitable path that CF follows. Fight referred to the regimented daily treatment which is required to stave off deterioration in physical health but also the fight for a normal and accepted life within education, work and social circumstances. Related to this was the theme surrounding form which reflected participants' feelings towards their own body and physical appearance (Jessup & Parkinson, 2010). Similarities to these themes can be found in the discussion forum post themes of daily CF treatment, progression of illness,

the future and psychosocial processes and issues. More specifically, the sub-themes of fear of the future, feeling different, body image, transitions and adjustment arising from the forum data reflect those themes raised in Jessup and Parkinson's (2010) study. Furthermore, similarities can be found with flight, described by the authors' as physical flight to different CF centres, imagined situations of being 'fine' and a desire to be 'anywhere but here' (Jessup & Parkinson, 2010). Equivalent topics are found in the current study in terms of the discussions between forum users about moving abroad and differences in CF healthcare, and also the recognition that some participant often presented to others that they were 'fine'. Finally, similarities were present to a lesser extent with Jessup and Parkinson's (2010) theme of philosophy. This referred to the meanings and metaphors held by each family to cope with the reality of CF, such as, 'That's life' or 'It was meant to be. Just do it' (Jessup & Parkinson, 2010). A minority of forum posts in the present study acknowledged that acceptance of CF and the realities it brings can help with coping but the use of metaphors or philosophies by participants did not emerge to same extent as in study by Jessup and Parkinson (2010).

The two more specifically focussed qualitative studies by Lowton (2004) and Macdonald (2006) also contain parallel themes to those topics discussed on the CF Trust online forum. Macdonald's (2006) study focussed on CF patients' experiences of going through the lung transplant process and the emergent themes were described as displacement, disorder, life in limbo and readjustment to wellness. Displacement refered to the process of realising and accepting that health had deteriorated to the point of requiring a transplant. This was followed by disordered emotions consisting of fear and hope for the future. At this point, participants described a need for support from those around them and from people who had been through the same process. Waiting for the telephone call notifying of a transplant was a difficult time for patients, involving some false alarms and disappointed hopes. Once transplant had been successful, participants required a period of adjustment to their new

health status and mixed feelings of guilt and gratitude for received another person's lungs (Macdonald, 2006). The themes described by Macdonald (2006) bear significant resemblance to the issues encapsulated by the themes of psychosocial processes and issues and progression of illness in the current study.

Similarities seen with Lowton's (2004) study are found within psychosocial issues but also in the themes of daily living and occupation and the future. This interview-based study focussed on exploring the experience of adults with CF in disclosing their diagnosis to others. The author described three different situations where disclosure may take place: low-, medium- and high-risk situations (Lowton, 2004). Disclosure within all these circumstances was influenced by the current health of participants, their physical appearance and self-identity and participants made active choices whether or not to reveal their CF. Low-risk situations were those involving casual encounters or meeting acquaintances. The emphasis for participants here was mostly on concealment of diagnosis but with awareness that coughing symptoms may give the diagnosis away. Medium-risk situations were those involving friends, where decisions to disclose their CF depended on the level of closeness and the perceived reaction of the other person. High-risk situations for disclosure were those involving potential partners or employers. Revelation of CF in each of these situations was considered as having the most significant impacts and potential consequences of all. CF would impact on the future of relationships due to the associated difficulties with fertility and the increased likelihood of an early death. In the case of employment, disclosure of CF at all stages of job application could have disadvantages (Lowton, 2004). All of these issues are represented within the discussion forum posts and it seems that forum users take the opportunity to discuss these issues with their CF peers.

In addition to the discussion topic themes noted within the forum posts, two additional observations were made about the general atmosphere of the posts and the individual

narratives of participants. Firstly, the majority of posts provided support and encouragement, information, advice or sharing of personal experiences. Those who shared feelings, experiences or concerns seemed to do so in order to illustrate a particular difficulty they were themselves facing or to help reassure or identify with another user's situation. Secondly, when examining posts of each individual user across themes, forum users seemed to seek or provide support differentially depending on their current needs and point of disease progression. Some of the information shared was of a personal nature and it would seem that participants felt that the forum was a safe place in which to reveal their difficulties and potential vulnerabilities. These observations suggest that a culture of sharing experiences and providing empathic responses has been built within this online community.

In relation to the survey questions focussing on internet resource use, it seems that the themes emerging from qualitative analysis of posts are comparable to the responses given for how people use the CF Trust forum. The top three reasons for using the CF Trust forum were to 1) find information about CF (96.9 per cent), 2) find out information about CF treatments (87.5 per cent) and 3) get health advice (75.0 per cent). When asked to rate the reasons for using the CF Trust Facebook website, it became apparent that this site is used differently to the CF Trust discussion forum. The most commonly identified uses for the Facebook site were 1) find out information about CF (50.0 per cent), 2) pass the time when in hospital (47.5 per cent), and 3) find out about CF events (45.5 per cent). Although participants used both sites to find out information about CF, a smaller proportion of participants identified this as a reason they used the Facebook page. The differential use of the CF Trust forum and Facebook site may be due to their differing contexts. Facebook is a widely-used social networking site targeted at the general public, whereas the CF Trust discussion forum is targeted specifically at those people whose lives have been affected by CF. Therefore, people may visit the CF Trust discussion forum when they have more specific questions or needs concerning their CF which their wider social network may not be able to adequately answer

or fully understand. On the other hand, Facebook provides a wider social connection with a greater variety of options to pass the time, such as sharing photographs or arranging events. Discussing more serious CF-related concerns in the public forum provided by Facebook may not feel as comfortable or beneficial as interactions which take place on the CF Trust discussion forum. Moreover, the additional comments made by participants identified the value of having shared experiences and understanding with other forum members. It seemed to provide people with a way to interact with others with whom they identified and with whom it would not be possible to meet face-to-face. Furthermore, involvement in the CF Trust forum allowed people with CF to offer advice and support others with CF.

When considering the aspects least liked about the CF Trust forum in comparison to the qualitative forum data analysis there were some differences. Survey participants frequently mentioned problems with arguments arising within forum threads or some users being insensitive, rude or antagonistic. However there was little representation of this within the forum posts which were analysed in Study 2. A handful of posts were noted to take a stance attempting to diffuse potential arguments but feelings of antagonism and hostility were not picked up as a key aspect of any posts per se. The lack of presence of arguments within the sample of posts used may be a result of the self-selecting sampling method. It is possible that research participants choosing to provide their username for inclusion within the qualitative aspect of the study were those users who made use of the forum more frequently or perhaps those who used it in a more positive manner.

The results of the qualitative analysis and summaries of the survey questions relating to forum use reflect similar findings to those within the existing literature on the use of online discussion forums or internet support groups (ISGs) for physical illness and mental health difficulties. Research has suggested a negative correlation between higher levels of social support or functioning and mental health symptoms (Anderson *et al.*, 2001; Burker *et al.*,

2000; Casier *et al.*, 2011; Havermans *et al.*, 2008; Riekert *et al.*, 2007; Szyndler *et al.*, 2005) and recent papers have shown that online interactions can meet social support needs of some populations. The literature focussing on physical health conditions other than CF has illustrated similar characteristics of mutual support, advice and information sharing within a range of private, public and purposeful online environments: breast cancer, arthritis, fibromyalgia (van Uden-Kraan *et al.*, 2008); depression (Takahashi *et al.*, 2009); polycystic ovary syndrome (Percy & Murray, 2010); and multiple diagnoses (Sanders, Rogers, Gardner, & Kennedy, 2011). To the author's knowledge, the only study which specifically focuses on internet support group (ISG) for people with CF is that described in Barber's (2008) thesis. This study investigated the impact of ISGs on patients with CF and found that patients' social needs could be met via online relationships and that these helped to overcome restrictions of segregation policies.

7.4 LIMITATIONS

During the process of completing this project, a number of limitations came to the attention of the researcher. The author acknowledges that the use of self-selected sample may mean that data are not fully representative of the entire target population. The sample is likely to include those users who are the most frequent visitors to the discussion boards and those who were most willing to take part (van Uden-Kraan *et al.*, 2008). The proportion of the forum users who chose not to participate may have felt that they did not have time to complete the survey or they may have been deterred by some aspect of the project, such as the inclusion of mental health measures or the use of their forum posts. Furthermore, it has been noted that more females take part in online research (Rhodes *et al.*, 2003) and this was apparent within the current research.

A second limitation is found within the demographic questionnaire. Participants were asked to report their most recent FEV_1% predicted reading and the accuracy of this can not be

guaranteed. Some CF patients may have several months between clinic visits where lung function tests are carried out and their FEV_1% reading may have changed since their last assessment. Also, saliency of the information may have played a role in remembering or knowing FEV_1% predicted score. For example, some clinicians may only routinely tell patients the outcome of their lung function test if it markedly low or patients may more easily remember their reading if it is particularly low or high. In defence of this potential limitation, it was seen that the average lung function fell in the region of previous studies of CF patients drawn from clinical settings and not from online populations (Havermans *et al.*, 2008; Riekert *et al.*, 2007; Sawicki *et al.*, 2011).

A third and final limitation is found within the online survey and choice of mental health measure. The CORE-OM 34 was selected for its consideration of a wide range of symptoms rather than a focus on only one subset of symptoms, e.g. depression or anxiety. However, many other studies within CF make use of the HADS, BDI or SCL-90-R and this makes direct comparisons with other studies more difficult. Despite this, the ability to present data in terms of the proportion of participants who scored above recommended clinical thresholds enabled some level of comparison with other studies.

Although not considered a limitation per se, it is inevitable within qualitative methodologies that the researcher involved will bring her own approach and background to the data analysis process. Throughout all stages of the analysis, the researcher attempted to remain close to the original data and avoid abstract interpretations. Coming from a background of clinical psychology, it is possible that this had some influence on the themes identified and synthesis of the data. However, in using a structured framework approach to the analysis, it is felt that any bias would have been minimised (Ritchie & Lewis, 2003; Smith & Firth, 2011). Furthermore, the researcher remained reflective throughout the analysis process, paid attention to contradictory posts and contrasted themes identified within forum posts with

those from the qualitative comments made in the online survey and previous research (Mays & Pope, 2000). These factors, alongside using an explicit and transparent analytical process, are known to improve the quality of qualitative research (Baym, 2009; Mays & Pope, 2000; Miles & Huberman, 1994; Ritchie & Lewis, 2003).

7.5 ETHICAL CONSIDERATION OF ONLINE RESEARCH

When embarking on this study using online data, a number of ethical considerations were taken into account. There has been considerable debate in recent years regarding the privacy of information contained on websites (Kozinets, 2010; Sveningsson Elm, 2009). Some researchers take the stance that information within the public domain, which can be accessed freely and is not password-protected, can be used in research without seeking prior consent from the authors. However, another branch of thought takes a more conservative approach and acknowledges that online community members may assume some level of privacy even though forums are in public view (Sveningsson Elm, 2009). Similarly, some web-users may not fully appreciate the open availability of the information they post and may not expect posts to be used in research (Corbyn, 2008; Kozinets, 2010; Sveningsson Elm, 2009). In the context of this study it was felt that a conservative approach was most ethical in terms of the use of online discussion forum data. Forum data was collected only from those survey participants who provided their forum user names and who consented to take part in the study. All participants were provided with full information about how data would be used, including that direct quotes may appear in research summaries and articles.

A second important aspect to consider when carrying out any type of psychological research is to ensure minimisation of harm. Rhodes *et al.* (2003) highlight the difficulties of offering support and identifying needs of participants when data is collected online as opposed to face-to-face. It has been suggested that providing contacts of supportive resources at the end of the research fails to be communicated to those participants who may exit the survey

before completion and subsequently miss the resource information. In order to remedy this problem, reference was made within the information sheet that participants may find some items within the survey distressing. If this was the case, participants were encouraged to talk with a member of their health team or general practitioner. At the end of the survey, participants were also provided with the contact details for a number of mental health and support resources should they require them.

A third and final point which was considered within the design and execution of the project was the matter of anonymity and confidentiality of data. This issue has received a great deal of attention in recent summaries of the ethical practice in online research (Kozinets, 2010; Sveningsson Elm, 2009). In terms of the quantitative survey, this was hosted by SurveyMonkey and once the closing date of the study had passed, responses were downloaded and deleted from this website. No IP addresses were collected within the survey and, in order to keep survey responses anonymous, usernames were separated from survey data once downloaded. Participants were asked not to provide any personally identifiable information, such as real name, address or email. Those completing the online survey were asked to give their date of birth (in order to ascertain age and also to provide a method of checking whether participants had completed the survey multiple times). In respect of the qualitative study, participants were asked to provide their CF Trust forum username if consenting to take part in this section of the project and for their forum posts to be used in the research. The CF Trust provides guidance for using their forum and advises users not to reveal any personal information including postal address, telephone number or email address. However, it has been acknowledged in the internet research literature that members of online communities sometimes post information which links pseudonym usernames to real names or circumstances (Kozinets, 2010). Using direct quotes within written reports of internet research projects has the danger of revealing the identity of research participants as powerful internet search engines could locate the original posting and its author's username.

As a consequence, the researcher felt that it was extremely important for participants to be informed that direct quotes may be used within the written reports and that they were aware that in consenting to take part in the study, direct quotes may be used. It was felt by the researcher that the information provided to participants and the inclusion of a consent question adequately addressed ethical considerations within this study.

7.6 CLINICAL IMPLICATIONS

Despite the ongoing debate about whether adults with CF experience elevated levels of mental health difficulty compared to the general population, the impacts of difficulties where they do occur can have wide-ranging implications for quality of life and treatment adherence (Cruz *et al.*, 2009; Quittner *et al.*, 2008). The Department of Health (2005) has recognised the importance of establishing peer support networks for people with long term conditions in order to help support self-management of illness. With the segregation policies in place for CF care, patients with CF are not able to do this face-to-face and, instead, have to utilise other technologies to build relationships with their CF peers. Online discussion forums and support groups present an opportunity for people with CF to access peer support and these resources are becoming more widely recognised as a viable option (Hasenecz, 2010).

A recent report resulting from collaboration between the Scottish Government and the Long Term Conditions Alliance Scotland (2011) highlighted that people living with long term conditions may not be receiving enough emotional and psychological support. They recommend that health services work jointly with third sector organisations to help fill this gap and the use of online communities may be one way of doing this. More specifically, the recognition that adults with CF may have unmet psychosocial needs is increasing and recommendations for CF services include the provision of a clinical psychologist (Oxley & Webb, 2005).

Research exploring the relative benefits of involvement in online support groups or communities is still in its infancy in terms of the CF population. However, there does appear to be some emerging support for a positive influence of online support on psychological wellbeing (Barber, 2008). Literature focussing on the experiences of people with other health conditions has shown mixed results. It seems that although some studies claim that involvement in online support groups leads to reduced depressive symptoms, the often poor quality of study designs, such as lack of a control condition or providing other potentially influential interventions at the same time, compromises the confidence in findings (Griffiths *et al.*, 2009).

The current project did not aim to assess the impact of involvement with an online community on mental health or quality of life. Instead, its focus was on finding out the mental health needs of this population and exploring the ways in which the discussion forums are used and the topics which arose. It was seen within this project that adults with CF value their online community and use it to access information, advice and support from peers who can truly identify with their predicament. It was apparent that people used the forum in different ways depending on their current life or health circumstances and, in line with similar research, it seems that online communities can help to meet social and emotional support needs for this population (Barber, 2008; van Uden-Kraan *et al.*, 2008).

In addition to the less formal provision of social and emotional support for people with CF, there may be a number of ways in which the online environment could be utilised for mental health and wellbeing services. The format of online discussion forums could be harnessed on a smaller, local scale and used to run psychoeducation, treatment adherence groups or psychotherapeutic groups without the risk of cross-infection. Video-conferencing facilities or web-cast links may be useful in promoting such contacts. An initial step towards making use of video-conferencing technology in CF care has been made at Nottingham City Hospital.

Fundraising efforts were successful in raising enough money to install video-conferencing facilities onto their CF ward to allow inpatients to communicate with each other and their families at home without the risks of cross-infection. It will be interesting to see whether this innovative introduction to inpatient care and its benefits to patients will be evaluated.

7.7 DIRECTIONS FOR FUTURE RESEARCH

This study of adults with CF who make use of online resources and social networking contributes to the current literature surrounding the mental health needs and wellbeing of adults with CF. It adds to this literature by explicitly considering self-esteem and its relationship with mental health symptoms and HRQoL. Furthermore, it provides an insight into the topics arising for discussion on a CF online forum. Inevitably, carrying out research and considering its findings within the context of published works leads to the development of further research questions. As outlined above, there are a number of possible clinical implications of this research, each with related avenues for research. In light of the discrepancies between estimates of mental health difficulties in the CF population, it may be worthwhile investigating the factors which influence resilience in the face of the challenges CF brings. This may include building on the work of Casier *et al.* (2008, 2011) and their focus on the role of acceptance in wellbeing of CF patients. Well-designed studies which take a longitudinal viewpoint of relevant factors may help disentangle the respective contributions of variables including gender, physical health status, social support, self-esteem, acceptance and psychological wellbeing. In retrospect, two additions could have been made to the current online survey which may be addressed in further research. Firstly, including a question asking participants to indicate whether or not they had received or were currently receiving input from psychology services would have been beneficial. This would have allowed calculation of the proportion of the mental health needs identified in this population which were unmet. Secondly, the use of a measure of perceived social support, such as the Significant Others Scale (Power *et al.*, 1988) would have provided an insight into

the potential differences between social functioning as measured by the CFQ-R and social support. If distinctions are made between online and non-virtual relationships, exploring these would add to the understanding of how patients with CF perceive each type of support and the benefit, or disadvantages, provided.

The beneficial impacts of online social support groups on the psychological well-being of people with CF are currently unknown. In other areas of health, a review paper suggests that there may be emotional benefits of involvement in online social support (Griffiths *et al.*, 2009). However the authors recognise that existing research in the area has considerable limitations and is generally of poor quality (Griffiths *et al.*, 2009). Another paper acknowledged that there may also be the possibility of harm in being involved in online support networks (Takahashi *et al.*, 2009). Further research may wish to focus on improving the quality of research in these areas but also in exploring the most beneficial and suitable types of support, particularly for those who already experience some level of mental health difficulty. It is known that a current randomised control study is underway in the USA looking at whether providing young people with CF with a web-enable phone, the CFFone™, is beneficial for improving knowledge, treatment adherence and social support in youth with CF (Marciel *et al.*, 2010). This provides access to a specific website with an online 'chat' environment which is overseen by psychology clinicians for incorrect medical information or potential psychological difficulties. It is hoped that the findings of this study will provide further knowledge on whether online support for people with CF is beneficial.

For those patients with CF who do experience mental health problems, further research is required to ascertain the efficacy of psychological therapies and their most beneficial components. Research exploring the efficacy of common psychotherapeutic interventions is severely lacking in CF (Glasscoe & Quittner, 2008) and this needs to be addressed. Furthermore, segregation policies, physical health limitations and the distinct nature of CF

being a life-limiting and currently incurable disease may have impacts on the relevance for particular therapies. Traditional cognitive-behavioural therapy (CBT) approaches are now widely used across mental health services but often encourage patients to engage in increased activity and challenge unhelpful thought processes. However, patients with CF, especially those in the later stages of the disease, may be restricted by their physical functioning and the negative cognitions they experience may be related to realistic concerns about the future or current health situations. Therefore, it could be more relevant to explore options from other approaches such as acceptance and commitment therapy which has built an evidence-base for many problems stemming from both psychological and physical health problems (Kangas & McDonald, 2011; Pull, 2009). Finally, if group interventions are thought to provide advantages over and above individual psychotherapy, it is possible that new technologies such as video-conferencing may enable therapists to deliver interventions to groups of CF patients.

8 JOURNAL ARTICLE

TITLE: Self-esteem and its relationship to mental health and quality of life in adults with cystic fibrosis who use an online discussion forum

RUNNING TITLE: Mental health, quality of life and self-esteem in CF

AUTHORS:

Melanie Jane Platten*

Dumfries & Galloway NHS Health Board, Department of Psychological Services and Research, Nithbank, Dumfries, DG1 2SA.

Telephone: 01387 244495

Fax: 01387 244496

Email: m.platten@nhs.net

Emily Newman

School of Health in Social Sciences, University of Edinburgh, Medical School, Teviot Place, Edinburgh, EH8 9AG

Telephone: 0131 651 3945

Email: emily.newman@ed.ac.uk

Ethel Quayle

School of Health in Social Sciences, University of Edinburgh, Medical School, Teviot Place, Edinburgh, EH8 9AG

Telephone: 0131 650 4698

Email: ethel.quayle@ed.ac.uk

*Corresponding author

ABSTRACT

Background: Despite research from the general population indicating an important role for self-esteem and social support in mental health, limited research into these two factors exists in the cystic fibrosis (CF) literature. Access to face-to-face CF peer support is limited due to segregation policies and internet technologies are becoming more frequently used to overcome this barrier. This study aimed to explore the relationships between self-esteem, mental health and health-related quality of life (HRQoL) in adults with CF who make use of online social support.

Method: Seventy-four participants recruited via an online discussion forum completed assessments of mental health, self-esteem and HRQoL.

Results: Comparably high levels of self-esteem were found, but HRQoL was lower than previous research. Thirty per cent of participants scored within the clinical range for mental health difficulty. Significant correlations indicated that lower self-esteem was associated with poorer quality of life and greater mental health difficulty.

Conclusions: Results are discussed in relation to clinical implications and potential uses for internet technologies to promote socialisation.

Key words:

Cystic fibrosis; mental health; self-esteem; quality of life; online support; social functioning

INTRODUCTION

With the vast majority of people with cystic fibrosis (CF) now living into adulthood researchers have become increasingly interested in their long-term quality of life and psychological well-being. Studies have indicated that the CF population report high levels of quality of life (1) but that levels of mental health difficulty vary. Some studies suggest higher levels of depression and/or anxiety compared to the general population (2-4) whereas others maintain that levels of mental health symptoms are comparable to the general population (5).

The reasons for this variability continue to be explored by research and factors identified as influential include being female, older, unemployed and having more severe illness (2-4).

Two factors which have been shown to play an important role in the mental health of the general population are self-esteem and social support. However, these two variables have received relatively little attention within the CF literature. Self-esteem has been linked with better mental health and psychological wellbeing in the general population (6) and has been shown to play an important role in mediating the relationship between dysfunctional thoughts and depression in adult participants (7). To the authors' knowledge, only two papers have explicitly assessed self-esteem in adults with CF with both papers focusing primarily on body image (8, 9). A recent publication by Brucefors et al. (10) speculated that low levels of self-esteem may have contributed to the higher levels of social dysfunction found in their sample of adults with CF compared with healthy adults. However, there was no direct measurement of self-esteem within their project and, as yet, there appear to be no published studies which consider the relationship between self-esteem and mental health in adults with CF.

Social support is important for adaptation to stress and higher levels of support are related to lower levels of mental health difficulty in the general population (11). Peer relationships have been seen to be affected in chronic illnesses (12) and, with the use of segregation in CF services to prevent cross-infection of respiratory bacteria, it is important to consider how people with CF can build and maintain relationships with and access support from their CF peers (13). The evidence from CF research overall seems to indicate that higher levels of social support and better levels of social functioning are related to better coping (14), improved adherence to treatment (14) and fewer mental health difficulties (15). Researchers have suggested that future studies continue to examine the potential buffering effects of social support (4) and explore how new technologies may enhance peer interaction and

overcome the restrictions of segregation for people with CF (13). One option for keeping in contact with CF peers is to use the internet and social networking websites, where interactions can take place without the risks of cross-infection. Online resources are becoming increasingly popular (16) and may provide an additional means of peer support for people with CF. However, little research has been carried out in the area of online resources and the groups of CF patients who make use of specific resources available. As a starting point, the present study aimed to assess the mental health needs and HRQoL (including social functioning) of adults with CF who make use of online resources. In addition, it aimed to assess the levels of self-esteem within this group and explore the relationships of self-esteem with mental health and HRQoL. In line with previous research involving the general population and CF patients, the authors hypothesised that lower self-esteem would be related to poorer mental health and lower levels of quality of life. Levels of mental health difficulty were hypothesised to be similar to recent research in CF which has indicated a slight elevation in the prevalence of mental health difficulty when compared to the general population.

METHODS

DESIGN

This was a cross-sectional study of adults with cystic fibrosis who make use of online discussion forums and social networking. Participants completed three quantitative assessment measures and provided demographic information.

PARTICIPANTS

Participants were recruited through an online discussion forum and Facebook page hosted by the Cystic Fibrosis Trust (CF Trust), a UK-based charity. A total of 105 eligible participants consented to take part in the study. Surveys were fully completed by 74 participants.

Calculations were carried out to establish the size of correlation which could be confidently detected for a sample size of 74, an alpha level of .05 and 80 per cent statistical power. Using a two-tailed hypothesis, this revealed that analysis could detect correlations of r = .31, a medium effect size (17).

Key demographic details for full and partially completed surveys were compared using non-parametric chi-squared and Mann-Whitney U tests. No significant differences ($p>.05$) were found between full- and partial-completers on the demographic variables of age, FEV_1% predicted, gender, ethnicity or lung transplant. Demographic variables for participants completing a full set of survey data are presented in Table 1.

PROCEDURE

Recruitment was initiated via a message posted on the CF Trust online discussion forum by a member of the CF Trust communications team. This linked to an information page describing the study. After 3 and 6 weeks, the researcher replied to the initial thread to thank those who had already taken part and to bring the notice to the top of the discussion area. In addition, 6 weeks after the original post on the CF Trust discussion forum, the communications team placed a recruitment notice on the CF Trust Facebook page followed by a repeat post from the researcher 2 weeks later. In total, the survey was open for ten weeks.

The information page contained a web-link to the online survey, hosted by Survey Monkey (www.surveymonkey.com). Participants self-selected to take part in the research and were eligible if they were 16 years old or over and had a diagnosis of CF. Participants were asked to provide consent prior to completing the online survey. Those who chose not to consent were directed to the end of the survey and thanked for their time.

Table 1. Demographic and basic clinical information summary for full data sets

Demographic		**Clinical**	
Age (mean ± SD)	27.8 ± 9.2		
Gender (n, % female)	57, 77.0%	FEV_1% (mean ± SD)	61.7 ± 25.0
Ethnicity (n, % White)	72, 97.3%	Lung transplant (n, % yes)	5, 6.8%
Education (n, % Higher Ed)	45, 60.8%		
Marital Status (N = 74)	**n (%)**	Age at CF diagnosis (N = 74)	**n (%)**
Single/never married	28 (37.8)	*At birth*	19 (25.7)
With a partner	25 (33.8)	*<6 months old*	18 (24.3)
Married/civil partnership	20 (27.0)	*6 – 12 months old*	10 (13.5)
Widowed	1 (1.4)	*1 – 4 years old*	13 (17.5)
		5 – 11 years old	3 (4.1)
Education (N = 74)		*12 – 17 years old*	3 (4.1)
Some secondary school or less	1 (1.3)	*18 – 25 years old*	3 (4.1)
GCSEs/O-levels	11 (14.9)	*>25 years old*	5 (6.7)
A/AS-levels	17 (22.9)		
Other higher education	13 (17.6)	CF Severity, No Transplant (FEV_1%; N = 54)	
University degree	21 (28.4)	*Normal (FEV_1% ≥90%)*	6 (11.1)
Professional or postgraduate	11 (14.9)	*Mild (70 – 89%)*	13 (24.1)
		Moderate (40 - 69%)	21 (38.9)
Work Status (N = 61)		*Severe (<40%)*	14 (25.9)
Working full- or part-time	32 (52.5)		
Not attending school or work due to health	12 (19.7)	CF Severity, Transplant (FEV_1%; N = 4)	
Attending school	11 (18.0)	*Normal (FEV_1% ≥90%)*	3 (75.0)
Seeking work	2 (3.3)	*Moderate (40 - 69%)*	1 (25.0)
Not working for other reasons	2 (3.3)		
Taking education courses at home	1 (1.6)		
Full-time homemaker	1 (1.6)		

MEASURES

Cystic Fibrosis Questionnaire – Revised (CFQ-R)

The CFQ-R (18, 19) is a CF-specific measure of health-related quality of life (HRQoL). The 14 years and over teen/adult UK version was used in this instance. The measure consists of 50 items which assess HRQoL within 9 domains. Previous research advocates the consideration of four core domains when studying HRQoL (15, 20) and, as a result, it was decided to include the CFQ-R subscales of physical functioning (eight items), emotional functioning (five items), role functioning (four items) and social functioning (six items). Each item on the CFQ-R is rated on a four-point Likert scale. A standardised score ranging between 0 – 100 is obtained for each subscale, with higher scores indicating better functioning. The CFQ-R teen/adult version is well established, valid and reliable (21). Cronbach's alpha internal consistency analyses were carried out for each of the four subscales: physical α=.96, emotional α=.82, role α=.86 and social α=.63.

Clinical Outcomes in Routine Evaluation – Outcome Measure 34 (CORE-OM)

The CORE-OM (22) is a 34-item self-report measure used to assess psychological and emotional distress. A total score can be calculated in addition to scores for four sub-domains: subjective well-being, functioning, symptoms and risk. Each item is rated on a four-point scale from 0 (not at all) to 4 (most or all of the time). Higher scores indicate greater difficulty. The measure has been shown to be reliable, valid and able to distinguish between clinical and non-clinical samples in the general population (23). CORE-OM total scores in the current sample had good internal consistency, Cronbach's α=.96.

Rosenberg self-esteem scale (24)

This 10-item self-report questionnaire is designed to measure global self-esteem. Each item is rated on a four-point scale (strongly disagree, disagree, agree, strongly agree) and results in a total score ranging from 10 – 40. Higher scores represent higher levels of self-esteem.

This measure has been shown to have satisfactory psychometric properties (25) and has been used in CF research (9). The current data had good internal consistency, Cronbach's α=.94.

Demographic questionnaire

Demographic data including age, gender, occupation, diagnosis and current physical health status (including most recent FEV_1% predicted reading and lung transplant status) were collected. FEV_1% predicted is a measure of lung function frequently used to classify severity of respiratory disease.

DATA ANALYSIS

Raw data were analysed using SPSS version 17. Scores for those participants who had fully completed the online survey (N = 74) were included in analyses. Kolmogorov-Smirnov statistical tests determined the normality of data distribution for FEV_1% predicted and each outcome measure variable. Data was normally distributed for all variables ($p>.05$) except for the CFQ-R subscale scores for physical functioning ($D = 0.17$, $p = .03$)[2].

Past research has suggested gender differences in HRQoL and psychological well-being in CF (26). Due to the unequal proportions of males and females in the current study, t-test comparisons of male and female mean scores on each outcome measure and FEV_1% predicted were carried out.

Correlations

Pearson's correlations were conducted between FEV_1% predicted and the three standardised outcome measures. Consideration was given to removing data of participants who had

[2] The researcher explored the benefit of transforming data using a square-root function. This succeeded in distributing scores normally ($D = 0.14$, $p = .11$). However, as the transformed scores did not result in any differences in the significance of subsequent statistical analyses compared to when using raw scores, it was decided to use the raw data for ease of interpretation of findings.

undergone a lung transplant. Given the small number of participants who were post-transplant (N = 5) and, as mean scores were not changed significantly when excluded, it was decided to include their data in subsequent analyses.

ETHICAL APPROVAL

Ethical approval was obtained from the School of Health in Social Science, University of Edinburgh research ethics panel. Collaboration with the CF Trust aided the development of the project and research information page.

RESULTS

DESCRIPTIVE STATISTICS

Table 2 presents the mean and standard deviation (SD) for each measure completed within the online survey. Self-esteem scores produced a mean of 29.9 (SD 7.1). The mean score on

Table 2. Means and standard deviations of outcome measure scores

Outcome measures		Mean scores (SD)		
		Male	Female	Total
FEV_1% predicted		66.73 (26.9)	60.47 (24.6)	61.66 (25.0)
CFQ-R	*Physical functioning*	58.82 (37.6)	43.42 (30.3)	46.96 (32.5)
	Role functioning	71.57 (23.8)	63.73 (23.2)	65.61 (23.4)
	*Emotional functioning***	71.76 (20.9)	52.98 (24.2)	57.30 (24.6)
	*Social functioning***	70.59 (15.6)	55.85 (20.5)	59.23 (20.4)
CORE-OM total score*		0.64 (0.5)	1.13 (0.7)	1.01 (0.7)
Rosenberg self-esteem scale**		33.94 (5.1)	28.68 (7.2)	29.89 (7.1)

** gender difference significant at the level of p<.05*

*** gender difference significant at the level of p<.01*

the CORE-OM was 1.01 (SD 0.7) and when classified according to clinical thresholds, 29.7 per cent (N = 22) of participants were within clinical range of symptoms. Average scores on the CFQ-R subscales ranged between 46.96 (SD 32.5) for physical functioning and 65.61 (SD 23.4) for role functioning. Mean scores of 57.30 (SD 24.6) and 59.23 (SD 20.4) were obtained for emotional and social functioning respectively.

CORRELATIONS

The results of the Pearson's correlation analyses are presented in Table 3. Significant positive correlations were found between the four subscales of the CFQ-R. Correlations between CFQ-R subscales and CORE-OM total score were all negative, with lower HRQoL scores associated with higher levels of mental health difficulty. Significant positive correlations between CFQ-R subscales and self-esteem scores were also found, indicating that better HRQoL is related to higher levels of self-esteem. Higher self-esteem scores were also associated with lower levels of mental health difficulty. Disease severity correlated positively with CFQ-R subscales of physical, role and social functioning. This suggests that those with better lung function experience better quality of life in these domains. The only non-significant correlations were found between disease severity and emotional functioning, CORE-OM total score and self-esteem.

GENDER DIFFERENCES

Exploratory two-tailed t-tests were carried out to compare scores of males (N = 17) and females (N = 57) on each measure. Mean scores, standard deviations and significance can be found in Table 2. Females scored significantly lower than males on the CFQ-R domains of emotional functioning ($t = 2.897$, $p = .005$) and social functioning ($t = 2.730$; $p = .008$). They were also seen to have lower levels of self-esteem ($t = 2.813$; $p = .006$) and higher total CORE-OM scores ($t = 2.534$; $p = .013$). No significant gender differences in FEV_1%

Table 3. Correlation coefficients and significance of online survey measure scores

		1	2	3	4	5	6	7
1. FEV_1% Predicted	r	1						
	N	58						
2. Physical functioning	r	.759*	1					
	N	58	74					
3. Role functioning	r	.445*	.757*	1				
	N	55	71	71				
4. Emotional functioning	r	.232	.569*	.686*	1			
	N	58	74	71	74			
5. Social functioning	r	.517*	.722*	.713*	.632*	1		
	N	58	74	71	74	74		
6. CORE-OM total score	r	-.135	-.464*	-.587*	-.841*	-.528*	1	
	N	58	74	71	74	74	74	
7. Rosenberg Self-esteem score	r	.233	.482*	.609*	.733*	.589*	-.827*	1
	N	58	74	71	74	74	74	74

* *Correlation is significant at the .01 level (two-tailed)*

predicted (t = 0.746; n.s.), physical functioning (t = 1.739; n.s.) or role functioning (t = 1.208; n.s.) were found.

DISCUSSION

Results showed that participants had good levels of self-esteem which compared well to previous CF participants and healthy controls (9). Mental health symptoms were at clinical levels for 29.7 per cent of the sample, which is higher than would be expected within the general population (27). However, this finding is in line with other studies indicating elevated mental health difficulties in the CF population (2-4). The results indicate that participants had variable levels of HRQoL depending on domain. Lowest scores were obtained in physical functioning whilst highest score were seen in role functioning. Role functioning refers to how much CF interferes with attending and keeping up with school, work or daily living activities. It also assesses whether CF interferes with achieving goals in areas of school, work or personal life. Compared to the baseline assessment scores reported in a recent longitudinal study of HRQoL in adults with CF (28), domain scores within this study are considerably lower. Sawicki et al.'s (28) sample were of a similar mean age (26.9 years, SD 9.7) and severity (FEV_1% 61.0, SD 23.0) yet mean scores for the four CFQ-R domains were: physical 67.9 (SD 27.4), role 81.1 (SD 19.4), emotional 76.3 (SD 20.0) and social 71.6 (SD 17.9).

One potential reason for the lower overall mean levels of quality of life compared to other studies may be due to the over-representation of females within the current sample population. Prior research has established that females with CF tend to fair worse medically than males (29) and that there are gender differences in psychological wellbeing. Females have been shown to experience higher levels of anxiety (3, 4), lower self-esteem (9) and lower HRQoL (1). The

current study presents data supporting the fact that females with CF tend to face greater difficulties with self-esteem, mental health symptoms and HRQoL compared to males.

Correlation analyses between outcome variables revealed that higher self-esteem was related to better mental health and better HRQoL. The significant relationships between higher mental health difficulty and lower levels of quality of life in all domains, including social functioning, are consistent with the results of previous research (2, 15). In terms of self-esteem, there are few studies which have considered the role of this construct in relation to mental health and HRQoL in the CF population. The significant relationships found within this study mirror the association between mental health and self-esteem in the general population (6). In addition, as suggested by Brucefors et al. (10), it suggests that self-esteem may have a role to play in low levels of social functioning and psychological wellbeing of adults with CF.

Significant correlations were also found between severity of CF disease (as measured by FEV_1% predicted) and physical, role and social functioning quality of life domains. However, no significant relationship was found between FEV_1% predicted and emotional functioning, mental health difficulty or self-esteem. FEV_1% predicted is a clinical measure used to gauge level of lung functioning and, therefore, lower scores indicate more severe lung disease. It follows that those people with poorer lung functioning will have more limitations on their physical health, be less able to carry out some roles within families, relationships or employment and be less able to socialise with friends.

Limitations

As with all research studies a number of limitations can be highlighted. Firstly, it is recognised that participants were self-selected and the sample is therefore unlikely to be fully representative

of the target population. Secondly, participants were asked to self-report their most recent FEV_1% predicted score and the accuracy of this can not be guaranteed. However, it was seen that the average lung function fell in the region of previous studies of CF patients recruited from clinical settings (28). Thirdly, one of the difficulties with the use of correlation statistics is that causality cannot be inferred. Although there are clearly relationships between variables, the influential impacts between them cannot be defined. Fourthly, the CORE-OM 34 was selected for its consideration of a wide range of symptoms rather than a focus on only one subset of symptoms, e.g. depression or anxiety. However, existing studies within CF make use of differing measures, making direct comparisons more difficult. Despite this, data was presented as the proportion of participants who scored above recommended clinical thresholds which allowed comparison with other studies.

Clinical implications and future research

This study of adults with CF who make use of online resources and social networking contributes to the current literature surrounding the mental health needs and wellbeing of adults with CF. Its novel contribution is in the exploration of self-esteem and its relationship with mental health symptoms and HRQoL. The current study reveals that higher levels of self-esteem are associated with better mental health and quality of life in adults with CF. Future studies which take a longitudinal approach in assessing relevant factors in psychological wellbeing and mental health may help disentangle the respective contributions such variables as gender, physical health status, social support and self-esteem. Factors which influence resilience in the face of the challenges CF brings might also be worthy of research. This work may build on a recent study by Casier et al. (15) looking at the role of acceptance in wellbeing of CF patients.

Despite the ongoing inconsistencies in reports of the prevalence of mental health problems in adults with CF, the impacts of difficulties where they do occur can have wide-ranging implications for HRQoL and treatment adherence (2). Given the gender differences in levels of mental health difficulty and self-esteem in the present study, it may be useful for clinical psychologists working within CF services to remain particularly vigilant for difficulties in females. In addition, in light of the relationship between poorer self-esteem and greater mental health difficulty for both males and females, the wider health team could be supported by clinical psychologists to recognise signs of low self-esteem and early detection of mental health difficulty. Where clinical levels of distress are identified, psychological therapies may be a useful treatment intervention. Traditional cognitive behavioural approaches may prove efficacious in this task but direct focus on self-esteem may also be beneficial. Further empirical research assessing the efficacy of psychological therapies in CF is still required (30). The value of emotional, psychological and peer support for people with long-term physical health conditions has been recently recognised by Scottish Government (31). Key international researchers also recognise that in order to enhance opportunities for people with CF to develop peer-support networks, use of technology needs to be explored (13). Online discussion forums and support groups present an environment for people with CF to access peer support and these resources are becoming more widely recognised as a viable option (16). The current sample of participants with CF who used online resources experienced lower levels of HRQoL than reported in previous literature and greater mental health difficulty compared to the general population. This may indicate that online forums are used by those who are more impaired or isolated due to their CF compared to the average patient with CF who is recruited into research studies via clinic situations. Further good quality research is required in order to provide insight into whether this medium of support can be beneficial. A review of the literature examining the effects on depression of involvement with internet support groups showed promise in some

areas, particularly for people suffering with breast cancer, but highlighted the shortage of well-designed studies (32). In relation to CF, a current randomised control trial in the USA is examining the influence of CF-specific online support on perceived social support, adherence and CF knowledge (33). This will be a useful addition to the literature and may be followed by further research to clarify the efficacy of online support for people with CF.

REFERENCES

1. Abbott J. Health-related quality of life measurement in cystic fibrosis: advances and limitations. Chronic Respiratory Disease. 2009 February 1, 2009; 6(1): 31-41.
2. Quittner AL, Barker DH, Snell C, Grimley ME, Marciel K, Cruz I. Prevalence and impact of depression in cystic fibrosis. Current Opinion in Pulmonary Medicine. 2008; 14(6): 582-8.
3. Goldbeck L, Besier T, Hinz A, Singer S, Quittner AL, Group T. Depression in German patients with cystic fibrosis. Chest. 2010; 138(4): 929 - 36.
4. Modi AC, Driscoll KA, Montag-Leifling K, Acton JD. Screening for symptoms of depression and anxiety in adolescents and young adults with cystic fibrosis. Pediatric Pulmonology. 2011; 46(2): 153-9.
5. Anderson DL, Flume PA, Hardy KK. Psychological functioning of adults with cystic fibrosis. Chest. 2001; 119 (4): 1079-84.
6. Mann M, Hosman CMH, Schaalma HP, de Vries NK. Self-esteem in a broad-spectrum approach for mental health promotion. Health Education Research. 2004 August 1, 2004; 19(4): 357-72.
7. Simpson J, Hillman R, Crawford T. Self-esteem and self-disgust both mediate the relationships between dysfunctional cognitions and depressive symptoms. Motivation and Emotion. 2010; 34: 399 - 406.

8. Abbott J, Conway S, Etherington C, Fitzjohn J, Gee L, Morton A, et al. Perceived body image and eating behavior in young adults with cystic fibrosis and their health peers. Journal of Behavioral Medicine. 2000; 23(6): 501 - 17.
9. Abbott J, Morton A, Musson H, Conway S, Etherington C, Gee L, et al. Nutritional status, perceived body image and eating behaviours in adults with cystic fibrosis. Clinical Nutrition. 2007; 26(1): 91 - 9.
10. Brucefors AB, Hjelte L, Hochwälder J. Mental health and sense of coherence among Swedish adults with cystic fibrosis. Scandinavian Journal of Caring Sciences. 2011; 25(2): 365-72.
11. Hefner J, Eisenberg D. Social Support and Mental Health Among College Students. American Journal of Orthopsychiatry. 2009; 79(4): 491-9.
12. Forgeron PA, King S, Stinson JN, McGrath PJ, MacDonald AJ, Chambers CT. Social functioning and peer relationships in children and adolesents with chronic pain: A systematic review. Pain Research and Management: The Journal of the Canadian Pain Society. 2010; 15(1): 27 - 41.
13. Quittner AL, Barker D, Marciel K, Grimley ME. Cystic fibrosis: A model for drug discovery and patient care. In: Roberts MC, Steele RG, editors. Handbook of Pediatric Psychology. 4th ed: Guilford Press; 2009.
14. White T, Miller J, Smith GL, McMahon WM, White T, Miller J, et al. Adherence and psychopathology in children and adolescents with cystic fibrosis. European Child & Adolescent Psychiatry. 2009 Feb; 18(2): 96-104.
15. Casier A, Goubert L, Theunis M, Huse D, De Baets F, Matthys D, et al. Acceptance and well-being in adolescents and young adults with CF. Journal of Pediatric Psychology. 2011; 36(4): 476 - 87.

16. Hasenecz NM. Cystic fibrosis social work - support on a changing journey. Social Work Today. 2010; 10(1): 22 - 6.
17. Cohen J. Statistical power analysis for the behavioral sciences. 2nd ed: Lawrence Earlbaum Associates, Inc.; 1988.
18. Bryon M, Stramik. Cystic Fibrosis Questionnaire - Revised. English language version.: Forest Laboratories; 2005.
19. Quittner AL, Buu A, Watrous M, Davis MA. Cystic Fibrosis Questionnaire - Revised. Forest Laboratories; 2002.
20. Cella D. Quality of life. In: Holland JC, editor. Psychooncology. New York, Oxford: Oxford University Press; 1998. p. 1135 - 46.
21. Palermo TM, Long AC, Lewandowski AS, Drotar D, Quittner AL, Walker LS. Evidence-based assessment of health-related quality of life and functional impairment in pediatric psychology. Journal of Pediatric Psychology. 2008; 33(9): 983-96.
22. Evans C, Mellor-Clark J, Margison F, Barkham M, McGrath G, Connell J, et al. Clinical Outcomes in Routine Evaluation: The CORE-OM. Journal of Mental Health. 2000; 9: 247-55.
23. Evans C, Connell J, Barkham M, Margison F, McGrath G, Mellor-Clark J, et al. Towards a standardised brief outcome measure: psychometric properties and utility of the CORE--OM. The British Journal of Psychiatry. 2002 January 1, 2002; 180(1): 51-60.
24. Rosenberg M. Society and the adolescent self-image. Princeton, NJ: Princeton University Press; 1965.
25. Sinclair SJ, Blais MA, Gansler DA, Sandberg E, Bistis K, LoCicero A. Psychometric Properties of the Rosenberg Self-Esteem Scale: Overall and Across Demographic Groups Living Within the United States. Evaluation & the Health Professions. 2010 March 1, 2010; 33(1): 56-80.

26. Gee L, Abbott J, Conway SP, Etherington C, Webb AK. Quality of life in cystic fibrosis: the impact of gender, general health perceptions and disease severity. Journal of Cystic Fibrosis. 2003; 2(4): 206-13.
27. Singleton N, Bumpstead R, O'Brien M, Lee A, Meltzer H. Psychiatric morbidity among adults living in private households, 2000. In: Statistics OfN, editor. London: HMSO; 2001.
28. Sawicki GS, Rasouliyan L, McMullen AH, Wagener JS, McColley SA, Pasta DJ, et al. Longitudinal assessment of health-related quality of life in an observational cohort of patients with cystic fibrosis. Pediatric Pulmonology. 2011; 46(1): 36-44.
29. Nick JA, Chacon CS, Brayshaw SJ, Jones MC, Barboa CM, Clair CGS, et al. Effects of Gender and Age at Diagnosis on Disease Progression in Long-term Survivors of Cystic Fibrosis. Am J Respir Crit Care Med. 2010 September 1, 2010; 182(5): 614-26.
30. Glasscoe CA, Quittner AL. Psychological interventions for people with cystic fibrosis and their familes. Cochrane Databse of Systematic Reviews. 2008 (3): Art. No.: CD003148.
31. Government S, Scotland LTCA. Emotional support matters: emotional and psychological support needs of people with long term conditions. Glasgow: Scottish Government; 2011.
32. Griffiths KM, Calear AL, Banfield M. Systematic review on internet support groups (ISGs) and depression (1): Do ISGs reduce depressive symptoms. Journal of Medical Internet Research. 2009; 11(3): 40.
33. Marciel KK, Saiman L, Quittell LM, Dawkins K, Quittner AL. Cell phone intervention to improve adherence: Cystic fibrosis care team, patient, and parent perspectives. Pediatric Pulmonology. 2010; 45(2): 157-64.

9 REFERENCES

Abbott, J. (2009). Health-related quality of life measurement in cystic fibrosis: advances and limitations. *Chronic Respiratory Disease, 6* (1), 31-41.

Abbott, J., Conway, S., Etherington, C., Fitzjohn, J., Gee, L., Morton, A. (2000). Perceived body image and eating behavior in young adults with cystic fibrosis and their health peers. *Journal of Behavioral Medicine, 23* (6), 501 - 517.

Abbott, J., Hart, A., Morton, A., Gee, L., & Conway, S. (2008). Health-related quality of life in adults with cystic fibrosis: The role of coping. *Journal of psychosomatic research, 64* (2), 149-157.

Abbott, J., Morton, A., Musson, H., Conway, S., Etherington, C., Gee, L. (2007). Nutritional status, perceived body image and eating behaviours in adults with cystic fibrosis. *Clinical Nutrition, 26* (1), 91 - 99.

Anderson, D.L., Flume, P.A., & Hardy, K.K. (2001). Psychological functioning of adults with cystic fibrosis. *Chest, 119 (4)*, 1079-1084.

Arrington-Sanders, R., Yi, M.S., Tsevat, J., Wilmott, R.W., Mrus, J.M., & Britto, M.T. (2006). Gender differences in health-related quality of life of adolescents with cystic fibrosis. *Health and Quality of Life Outcomes, 4* (5).

Barber, J.S. (2008). *E-patients and caregivers coping with cystic fibrosis: the relationship between relational satisfaction and attitudes toward groups, loneliness, and social support online.* Master of Arts, University of Akron, Akron.

Barker, D.H. (2010). *Measurement of disease specific social support in adolescents with cystic fibrosis.* Doctor of Philosophy, University of Miami.

Barker, C., Pistrang, N., & Elliot, R. (2002). *Research methods in clinical psychology: an introduction for students and practitioners* (2nd ed.). Chichester, England.: John Wiley & Sons, Ltd.

Baym, N.K. (2009). What constitutes quality in qualitative internet research? In A. N. Markham & N. K. Baym (Eds.), *Internet inquiry: conversations about method* (pp. 173 - 189): SAGE Publications, Inc.

Beck, A.T., Steer, R.A., & Barbin, M.G. (1988). Psychometric properties of the Beck Depression Inventory: twenty-five years of evaluation. *Clinical Psychology Review, 8*, 77 - 100.

Bregnballe, V., Thastum, M., & Schiøtz, P.O. (2007). Psychosocial problems in children with cystic fibrosis. *Acta Pædiatrica, 96* (1), 58-61.

Brucefors, A.B., Hjelte, L., & Hochwälder, J. (2011). Mental health and sense of coherence among Swedish adults with cystic fibrosis. *Scandinavian Journal of Caring Sciences, 25* (2), 365-372.

Bryon, M., & Stramik. (2005). *Cystic Fibrosis Questionnaire - Revised. English language version.*: Forest Laboratories.

Burker, E.J., Carels, R.A., Thompson, L.F., Rodgers, L., & Egan, T. (2000). Quality of life in patients awaiting lung transplant: Cystic fibrosis versus other end-stage lung diseases. *Pediatric Pulmonology, 30 (6)*, 453-460.

Burker, E.J., Sedway, J., Carone, S., Burker, E.J., Sedway, J., & Carone, S. (2004). Psychological and educational factors: better predictors of work status than FEV1 in adults with cystic fibrosis. [Research Support, Non-U.S. Gov't]. *Pediatric Pulmonology, 38* (5), 413-418.

Casier, A., Goubert, L., Huse, D., Theunis, M., Franckx, H., Robberecht, E. (2008). The role of acceptance in psychological functioning in adolescents with cystic fibrosis: A preliminary study. *Psychology & Health, 23* (5), 629-638.

Casier, A., Goubert, L., Theunis, M., Huse, D., De Baets, F., Matthys, D. (2011). Acceptance and well-being in adolescents and young adults with CF. *Journal of Pediatric Psychology, 36* (4), 476 - 487.

Cella, D. (1998). Quality of life. In J. C. Holland (Ed.), *Psychooncology* (pp. 1135 - 1146). New York, Oxford: Oxford University Press.

Cohen, J. (1988). *Statistical power analysis for the behavioral sciences* (2nd ed.): Lawrence Earlbaum Associates, Inc.

Cohen, S., & Wills, T. (1985). Stress, social support, and the buffering hypothesis. *Psychological Bulletin, 98* (2), 310-357.

Corbyn, Z. (2008). MySpace and your research: will red tape hit web data use? *The Times Higher Education Supplement*. Retrieved from http://www.timeshighereducation.co.uk/story.asp?storyCode=402665&sectioncode=26

Coyne, J.C., & Downey, G. (1991). Social Factors and Psychopathology: Stress, Social Support, and Coping Processes. *Annual Review of Psychology, 42* (1), 401-425.

Cruz, I., Marciel, K.K., Quittner, A.L., & Schechter, M.S. (2009). Anxiety and Depression in Cystic Fibrosis. *Semin Respir Crit Care Med, 30* (05), 569 - 578.

Cystic Fibrosis Trust (2011). *About cystic fibrosis.* Retrieved 3rd March 2011, from www.cftrust.org.uk/aboutcf

Department of Health (2005). *Supporting people with long term conditions: an NHS and social care model to support local innovations and integration*. Leeds, UK: COI for the Department of Health.

Delelis, G., Christophe, V., Leroy, S., Vanneste, J., & Wallaert, B. (2008). The effects of cystic fibrosis on couples: Marital satisfaction, emotions, and coping strategies. *Scandinavian Journal of Psychology, 49 (6)*, 583-589.

Durst, C.L., Horn, M.V., MacLaughlin, E.F., Bowman, C.M., Starnes, V.A., & Woo, M.S. (2001). Psychosocial responses of adolescent cystic fibrosis patients to lung transplantation. *Pediatric Transplantation, 5* (1), 27-31.

Ernst, M.M., Johnson, M.C., & Stark, L.J. (2010). Developmental and psychosocial issues in cystic fibrosis. *Child & Adolescent Psychiatric Clinics of North America, 19*, 263 - 283.

Evans, C., Connell, J., Barkham, M., Margison, F., McGrath, G., Mellor-Clark, J. (2002). Towards a standardised brief outcome measure: psychometric properties and utility of the CORE--OM. *The British Journal of Psychiatry, 180* (1), 51-60.

Evans, C., Mellor-Clark, J., Margison, F., Barkham, M., McGrath, G., Connell, J. (2000). Clinical Outcomes in Routine Evaluation: The CORE-OM. *Journal of Mental Health, 9*, 247-255.

Forgeron, P.A., King, S., Stinson, J.N., McGrath, P.J., MacDonald, A.J., & Chambers, C.T. (2010). Social functioning and peer relationships in children and adolesents with chronic pain: A systematic review. *Pain Research and Management: The Journal of the Canadian Pain Society, 15* (1), 27 - 41.

Geddes, D. (2001). Of isoates and isolation: Pseudomonas aeruginosa in adults with cystic fibrosis. *Lancet, 358*, 522 - 523.

Gee, L., Abbott, J., Conway, S.P., Etherington, C., & Webb, A.K. (2003). Quality of life in cystic fibrosis: the impact of gender, general health perceptions and disease severity. *Journal of Cystic Fibrosis, 2* (4), 206-213.

Gilbody, S., & Barkham, M. (2007). Diagnosing depression in primary care using self-completed instruments: UK validation of PHQ-9 and CORE-OM. *British Journal of General Practice, 57* (541), 650-652.

Glare, P., Pereira, G., Kristjanson, C. J., Stockler, M., & Tattersall, M. (2004). Systematic review of the efficacy of antiemetics in the treatment of nausea in patients with far-advanced cancer. *Support Care Cancer, 12*, 432 - 440.

Glasscoe, C.A., & Quittner, A.L. (2008). Psychological interventions for people with cystic fibrosis and their familes. *Cochrane Databse of Systematic Reviews* (3), Art. No.: CD003148.

Goldbeck, L., Besier, T., Hinz, A., Singer, S., Quittner, A.L., & Group, T. (2010). Depression in German patients with cystic fibrosis. *Chest, 138* (4), 929 - 936.

Griffiths, K.M., Calear, A.L., & Banfield, M. (2009). Systematic review on internet support groups (ISGs) and depression (1): Do ISGs reduce depressive symptoms. *Journal of Medical Internet Research, 11* (3), 40.

Hasenecz, N.M. (2010). Cystic fibrosis social work - support on a changing journey. *Social Work Today, 10* (1), 22 - 26.

Havermans, T., Colpaert, K., & Dupont, L.J. (2008). Quality of life in patients with Cystic Fibrosis: Association with anxiety and depression. *Journal of Cystic Fibrosis, 7* (6), 581-584.

Hefner, J., & Eisenberg, D. (2009). Social Support and Mental Health Among College Students. *American Journal of Orthopsychiatry, 79* (4), 491-499.

Herzer, M., Umfress, K., Aljadeff, G., Ghai, K., & Zakowski, S.G. (2009). Interactions with parents and friends among chronically Ill children: Examining social networks. *Journal of Developmental and Behavioral Pediatrics, 30 (6)*, 499-508.

Holahan, C.J., & Moos, R.H. (1981). Social support and psychological distress: a longitudinal analysis. *Journal of Abnormal Psychology, 90* (4), 365 - 370.

Ireys, H.T., Gross, S.S., Werthamer-Larsson, L.A., & Kolodner, K.B. (1994). Self-esteem of young adults with chronic health conditions: appraising the effects of perceived impact. *Journal of Developmental and Behavioral Pediatrics, 15* (6), 409-415.

Ishihara, L., & Brayne, C. (2005). A systematic review of nutritional risk factors in Parkinson's disease. *Nutritional Research Reviews, 18*, 259 - 282.

Jessup, M., & Parkinson, C. (2010). "All at Sea": The Experience of Living With Cystic Fibrosis. *Qualitative Health Research, 20* (3), 352-364.

Kangas, M., & McDonald, S. (2011). Is it time to act? The potential of acceptance and commitment therapy for psychological problems following acquired brain injury. *Neuropsychological Rehabilitation, 21* (2), 250 - 276.

Kinnear, P.R., & Gray, C.D. (2008). *SPSS 15 made simple*. East Sussex, England: Psychology Press.

Kozinets, R.V. (2010). Conducting ethical netography. *Netography: doing ethnographic research online* (pp. 136 - 156): SAGE Publications, Inc.

Lewis, J., & Ritchie, J. (2003). Generalising from qualitative research. In J. Ritchie & J. Lewis (Eds.), *Qualitative Research Practice* (pp. 263 - 286). London: Sage.

Liang, H. Y., Zhou, H., Li, X. L., Yin, Z. H., Guan, P., & Zhou, B. S. (2010). Chemo-radiotherapy for advanced non-small cell lung cancer: concurrent or sequential? It's no longer the question: a systematic review. *International Journal of Cancer, 127* (3), 718 - 728.

Liddle, J., Williamson, M., & Irwig, L. (1996). *Method for evaluating research and guideline evidence*. Sydney: New South Wales Department of Health.

Littlewood, J. (2004). *Looking back over 40 years and what the future holds.* Paper presented at the 27th European Cystic Fibrosis Conference, Birmingham.

Lowton, K. (2004). Only when I Cough? Adults' Disclosure of Cystic Fibrosis. *Qualitative Health Research, 14* (2), 167-186.

Macdonald, K. (2006). Living in limbo--patients with cystic fibrosis waiting for transplant. *British journal of nursing (Mark Allen Publishing), 15 (10)*, 566-572.

Mann, M., Hosman, C.M.H., Schaalma, H.P., & de Vries, N.K. (2004). Self-esteem in a broad-spectrum approach for mental health promotion. *Health Education Research, 19* (4), 357-372.

Marciel, K.K., Saiman, L., Quittell, L.M., Dawkins, K., & Quittner, A.L. (2010). Cell phone intervention to improve adherence: Cystic fibrosis care team, patient, and parent perspectives. *Pediatric Pulmonology, 45* (2), 157-164.

Masding, M.G., Ashley, K., & Klejdys, S. (2011). Introduction of a counselling service for patients with type 1 diabetes: better glycaemic control and reduced anxiety. *Practical Diabetes International, 28* (1), 28-30.

Mays, N., & Pope, C. (2000). Assessing quality in qualitative research. *BMJ, 320* (7226), 50-52.

Miles, M.B., & Huberman, A.M. (1994). *Qualitative data analysis: an expanded sourcebook* (2nd edition ed.). Thousand Oaks, Callifornia: SAGE Publucations.

Modi, A.C., Driscoll, K.A., Montag-Leifling, K., & Acton, J.D. (2011). Screening for symptoms of depression and anxiety in adolescents and young adults with cystic fibrosis. *Pediatric Pulmonology, 46* (2), 153-159.

Nick, J.A., Chacon, C.S., Brayshaw, S.J., Jones, M.C., Barboa, C.M., Clair, C.G.S. (2010). Effects of Gender and Age at Diagnosis on Disease Progression in Long-term Survivors of Cystic Fibrosis. *Am. J. Respir. Crit. Care Med., 182* (5), 614-626.

Oxley, H., & Webb, A.K. (2005). How a clinical psychologist manages the problems of adults with cystic fibrosis. *Journal of the Royal Society of Medicine, 98* (suppl. 45), 37 - 45.

Palermo, T.M., Long, A.C., Lewandowski, A.S., Drotar, D., Quittner, A.L., & Walker, L.S. (2008). Evidence-based assessment of health-related quality of life and functional impairment in pediatric psychology. *Journal of Pediatric Psychology, 33* (9), 983-996.

Pearson, D.A., Pumariega, A.J., & Seilheimer, D.K. (1991). The Development of Psychiatric Symptomatology in Patients with Cystic Fibrosis. *Journal of the American Academy of Child and Adolescent Psychiatry, 30* (2), 290-297.

Percy, C., & Murray, S. (2010). The role of an online peer-to-peer health community in addressing psychosocial concerns and social support in polycystic ovary syndrome. *International Journal of Web Based Communities, 6* (4), 349 - 361.

Pfeffer, P.E., Pfeffer, J.M., & Hodson, M.E. (2003). The psychosocial and psychiatric side of cystic fibrosis in adolescents and adults. [Review]. *Journal of Cystic Fibrosis, 2* (2), 61-68.

Power, M.J., Champion, L.A., & Aris, S.J. (1988). The development of a measure of a social support: the Significant Others Scale (SOS). *British Journal of Clinical Psychology, 27* (4), 349 - 358.

Public Health Resources Unit (2006). *10 questions to help you make sense of qualitative research*. England.

Pull, C.B. (2009). Current empirical status of acceptance and commitment therapy. *Current Opinion in Psychiatry, 22* (1), 55-60.

QSR International (2008). NVivo 8 (Version Student). Australia: QSR International.

Quittner, A.L., Barker, D., Marciel, K., & Grimley, M.E. (2009). Cystic fibrosis: A model for drug discovery and patient care. In M. C. Roberts & R. G. Steele (Eds.), *Handbook of Pediatric Psychology* (4th ed.): Guilford Press.

Quittner, A.L., Barker, D.H., Snell, C., Grimley, M.E., Marciel, K., & Cruz, I. (2008). Prevalence and impact of depression in cystic fibrosis. *Current Opinion in Pulmonary Medicine, 14* (6), 582-588.

Quittner, A.L., Buu, A., Watrous, M., & Davis, M.A. (2002). *Cystic Fibrosis Questionnaire - Revised*: Forest Laboratories.

Rhodes, S.D., Bowie, D.A., & Hergenrather, K.C. (2003). Collecting behavioural data using the world wide web: considerations for researchers. *Journal of Epidemiology and Community Health, 57*, 68 - 73.

Riekert, K.A., Bartlett, S.J., Boyle, M.P., Krishnan, J.A., & Rand, C.S. (2007). The association between depression, lung function, and health-related quality of life among adults with cystic fibrosis. *Chest, 132* (1), 231 - 237.

Ritchie, J., & Lewis, J. (2003). *Qualitative research practice: a guide for social science students and researchers*: SAGE.

Rosenberg, M. (1965). *Society and the adolescent self-image.* Princeton, NJ: Princeton University Press.

Russo, K., Donnelly, M., & Reid, A.J.M. (2006). Segregation--the perspectives of young patients and their parents. *Journal of Cystic Fibrosis, 5* (2), 93-99.

Sanders, C., Rogers, A., Gardner, C., & Kennedy, A. (2011). Managing 'difficult emotions' and family life: exploring insights and social support within online self-management training. *Chronic Illness, Published online.*

Sawicki, G.S., Rasouliyan, L., McMullen, A.H., Wagener, J.S., McColley, S.A., Pasta, D.J. (2011). Longitudinal assessment of health-related quality of life in an observational cohort of patients with cystic fibrosis. *Pediatric Pulmonology, 46* (1), 36-44.

Scottish Government & Long Term Conditions Alliance Scotland (2011). Emotional support matters: emotional and psychological support needs of people with long term conditions.

Glasgow: Scottish Government Retrieved from http://www.ltcas.org.uk/documents/LTCASEmotionalReportFinal.pdf.

Scottish Intercollegiate Guidelines Network (2008). *SIGN 50: A guideline developer's handbook*. (Revised ed.): SIGN, Edinburgh.

Simpson, J., Hillman, R. & Crawford, T. (2010). Self-esteem and self-disgust both mediate the relationship between dysfunctional cognitions and depressive symptoms. Motivation and Emotion, 34, 399-406.

Sinclair, S.J., Blais, M.A., Gansler, D.A., Sandberg, E., Bistis, K., & LoCicero, A. (2010). Psychometric Properties of the Rosenberg Self-Esteem Scale: Overall and Across Demographic Groups Living Within the United States. *Evaluation & the Health Professions, 33* (1), 56-80.

Singleton, N., Bumpstead, R., O'Brien, M., Lee, A., & Meltzer, H. (2001). *Psychiatric morbidity among adults living in private households, 2000.* London: HMSO.

Smith, J., & Firth, J. (2011). Qualitative data analysis: the framework approach. *Nurse Researcher, 18* (2), 52 - 62.

Soon, Y. Y., Stockler, M., Askie, L. M., & Bajer, M. J. (2009). Duration of chemotherapy for advanced non-small-cell lung cancer: a systematic review and meta-analysis of randomized trials. *Journal of Clinical Oncology, 27* (20), 3277 - 3283.

Spilker, B. (1996). Introduction. In B. Spilker (Ed.), *Quality of life and pharmacoeconomics in clinical trials* (pp. 1 - 10). Philadelphia, PA: Lippincott-Raven.

Srivastava, A. & Thomson, S. B. (2009). Framework analysis: a qualitative methodology for applied poicy research. *Journal of Administration and Governance, 4* (2), 72 - 79.

Stephenson, A., Hux, J., Tullis, E., Austin, P.C., Corey, M., & Ray, J. (2011). Higher risk of hospitalization among females with cystic fibrosis. *Journal of cystic fibrosis : official journal of the European Cystic Fibrosis Society, 10* (2), 93-99.

Sveningsson Elm, M. (2009). How do various notions of privacy influence decisions in qualitative internet research. In A. N. Markham & N. K. Baym (Eds.), *Internet inquiry: conversations about method* (pp. 69 - 87): SAGE Publications, Inc.

Szyndler, J.E., Towns, S.J., van Asperen, P.P., & McKay, K.O. (2005). Psychological and family functioning and quality of life in adolescents with cystic fibrosis. *Journal of Cystic Fibrosis, 4 (2)*, 135-144.

Takahashi, Y., Uchida, C., Miyaki, K., Sakai, M., Shimbo, T., & Nakayama, T. (2009). Potential benefits and hamrs of a peer support social network service on the internet for people with depressive tendencies: qualitative content analysis and social network analysis. *Journal of Medical Internet Research, 11* (3), 29.

van Uden-Kraan, C.F., Drossaert, C.H.C., Taal, E., Shaw, B.R., Seydel, E.R., & van de Laar, M.A.F.J. (2008). Empowering processes and outcomes of participation in online support groups for patients with breast cancer, arthritis, or fibromyalgia. *Qualitative Health Research, 18* (3), 405 - 417.

White, T., Miller, J., Smith, G.L., McMahon, W.M., White, T., Miller, J. (2009). Adherence and psychopathology in children and adolescents with cystic fibrosis. *European Child & Adolescent Psychiatry, 18* (2), 96-104.

World Health Organisation (2002). Services for adults with cystic fibrosis: report of a joint WHO/ICF(M)A/IACFA meeting. WHO. Last retrieved on 3rd October 2011 from http://www.cfww.org/docs/who/2002/services_for_adults_with_cystic_fibrosis.pdf.

Yardley, L. (2008). Chapter 11: Demonstrating validity in qualitative psychology. In J. A. Smith (Ed.), *Qualitative Psychology: A practical guide to reserach methods* (pp. 235 - 251). London: SAGE.

Zigmund, A.S., & Snaith, R.P. (1983). The Hospital Anxiety and Depression scale. *Acta Psychiatr Scand, 67*, 361 - 370.

10 APPENDICES

APPENDIX A: SYSTEMATIC REVIEW PROFORMAS

Critical Review and Data Extraction Proforma
[Adapted from SIGN 50 methodology checklists]

Paper details
Author:
Title:
Citation:
Type of publication:
Country of origin:
Source of funding:

Section 1: Internal validity

In a well conducted study...	***In this study the criterion is:***
1.1 The study addresses an appropriate and clearly focused question.	Well covered Adequately addressed Poorly addressed Not addressed Not reported Not applicable
SELECTION OF PARTICIPANTS	
1.2 The group(s) being studied is/are selected from appropriate source populations (and are comparable in all respects other than the factor under investigation).	Well covered Adequately addressed Poorly addressed Not addressed Not reported Not applicable
1.3 The study indicates how many of the people asked to take part and did so, in each of the groups being studied.	Well covered Adequately addressed Poorly addressed Not addressed Not reported Not applicable
1.4 What percentage of individuals or clusters recruited dropped out before the study was completed.	
1.5 Comparison is made between full participants and those who dropped out.	Well covered Adequately addressed Poorly addressed Not addressed Not reported Not applicable
ASSESSMENT	
1.6 The outcomes are clearly defined	Well covered Adequately addressed Poorly addressed Not addressed

	Not reported Not applicable
1.7 Evidence from other sources is used to demonstrate that the method of outcome assessment is valid and reliable.	Well covered Adequately addressed Poorly addressed Not addressed Not reported Not applicable
CONFOUNDING VARIABLES	
1.8 The main potential confounders are identified and taken into account in the design and analysis.	Well covered Adequately addressed Poorly addressed Not addressed Not reported Not applicable
STATISTICAL ANALYSIS	
1.9 Appropriate statistics were used.	Yes / No Type of analysis:
1.10 Statistical power was taken into account in analysis and conclusions.	Well covered Adequately addressed Poorly addressed Not addressed Not reported Not applicable
1.11 Appropriate adjustments were made for multiple comparisons.	Well covered Adequately addressed Poorly addressed Not addressed Not reported Not applicable
1.12 Authors report appropriate statistical figures e.g. p-values, z- or t-scores, correlation co-efficients etc.	Well covered Adequately addressed Poorly addressed Not addressed Not reported Not applicable
Section 2: OVERALL ASSESSMENT OF STUDY	
2.1 How well was the study done to minimise the risk of bias or confounding, and to establish a causal relationship between exposure and effect? Add up scores for each item in section 1	Total Score – Total possible score = 20 if all items applicable. (well covered = 2, adequately covered = 1, poorly addressed, not addressed, not reported = 0)
2.2 Taking into account clinical considerations, your evaluation of the methodology used, and the statistical power of the study, are you certain that the overall effect is due to the factors being	

investigated?	
SECTION 3: DATA EXTRACTION	
3.1 How many patients are included in this study?	
3.2 What are the main characteristics of the study population?	Age Gender Ethnicity Socio-economic status Disease characteristics Co-morbidities Hospital/community/urban/rural
3.3 How many centres are participants recruited from?	
3.4 What were the inclusion criteria?	
3.5 What were the exclusion criteria?	
3.6 What outcome measure(s) are used in the study? *List all outcomes that are used to assess the factors being examined in the study*	
3.7 Were data from participants who dropped out compared to those who continued?	Yes/No/Not applicable Number of participants who did not complete:
3.8 What were the outcomes of the measures used?	

Measure	n	Mean	Std Dev.
1.			
2.			
3.			
4.			
5.			
6.			

3.9 What comparisons are made in the study? *Are comparisons made between presence or absence of an environmental / prognostic factor, or different levels of the factor?*	
3.10 Did any of the comparisons/analyses reach statistical significance?	
3.11 Does this study help to answer your key question? *Summarise the main conclusions of the study and indicate how it relates to the key question?*	
3.12 Additional comments:	

Critical Review and Data Extraction Proforma – QUALITATIVE STUDIES
Critical Appraisal Skills Programme framework – Public Health Resources Unit, England, 2006

Paper details

Author:
Title:
Citation:
Type of publication:
Country of origin:
Source of funding:

1. Was there a clear statement of the aims of the research? What the goal was Why it is important Its relevance	
2. Is a qualitative methodology appropriate? If the research seeks to interpret or illuminate the actions and/or subjective experiences of research participants.	
Is it worth continuing?	
3. Was the research design appropriate to address the aims of the research? If the researcher has justified the research design (e.g. have they discussed how they decided which methods to use?)	
4. Was the recruitment strategy approrpaite to the aims of the research? If the researcher has explained how the participants were selected. If they explained why the participants they selected were the most appropriate to provide access to the type of knowledge sought by the study If there are any discussion around recruitment (e.g. why some people chose not to take part)	
5. Were the data collected in a way that addressed the research issue? If the setting for data collection was justified If it is clear how data were collected (e.g. focus group, semi-structured interview) If the researcher has justified the methods chosen If the researcher has made the methods explicit If methods were modified during the study. How and why? If the form of data is clear If the researcher has discussed saturation of data.	
6. Has the relationship between researcher and participants been adequately considered?	

If the research critically examined their own role, potential bias, and influence during formulation of research questions and data collection How research responded to events during the study and whether they considered the implications of any change in the research design	
7. Have ethical issues been taken into consideration? If there are sufficient details of how the research was explained to participants for the reader to assess whether ethical standards were maintained If the research has discussed issues raised by the study If approval has been sought from the ethics committee	
8. Was the data analysis sufficiently rigorous? If there is an in-depth description of the analysis process If thematic analysis is used, clear how the themes/cats were derived? Whether the researcher explains how the data presented were selected from the original sample to demonstrate the analysis process If sufficient data are presented to support the findings To what extent contradictory data are taken into account Whether the research critically examined their own role, potential bias and influence during analysis and selection of data for presentation	
9. Is there a clear statement of findings? If the findings are explicit If there is adequate discussion of the evidence both for and against the researchers arguments If the researcher has discussed the credibility of their findings If the findings are discussed in relation to the original research questions	
10. How valuable is the research? If the research discusses the contribution the study makes to existing knowledge or understanding If they identify new areas where research is necessary If the researchers have discussed whether or how the findings can be transferred to other populations or considered other ways the research may be used.	

SECTION 3: DATA EXTRACTION	
3.1 How many patients are included in this study?	
3.2 What are the main characteristics of the study population?	

3.3 How many centres are participants recruited from?	
3.4 What were the inclusion criteria?	
3.5 What were the exclusion criteria?	
3.6 What outcome measure(s) are used in the study? *List all outcomes that are used to assess the factors being examined in the study*	
3.7 What were the outcomes of the measures used?	
3.8 What comparisons are made in the study? *Are comparisons made between presence or absence of an environmental / prognostic factor, or different levels of the factor?*	
3.9 Did any of the comparisons/analyses reach statistical significance?	
3.10 Does this study help to answer your key question? *Summarise the main conclusions of the study and indicate how it relates to the key question?*	
3.11 Additional comments:	

APPENDIX B: ETHICS APPROVAL LETTER

School of Health in Social Science
Medical School, Teviot Place
Edinburgh
EH8 9AG

Telephone: 0131 651 3972
Fax: 0131 651 3971

11 January, 2010

Dear Melanie,

Re: Project Title: Cystic Fibrosis, psychological well-being and online social support

Your proposal has been reviewed and approved by the Programme Ethics Panel. The following comments are for your guidance only and should be discussed with your supervisor.

Good luck with the research,

Yours sincerely,

Ethel Quayle
Ethics Tutor

Ethics Approval Letter…. Cont'd

Researcher: Melanie Platten, Trainee Clinical Psychologist
Project Title: Cystic Fibrosis, psychological well-being and online social support

- The revised proposal is improved and Melanie has worked hard to address the comments. All of the points I raised have been dealt with satisfactorily. I am, however, unsure of the decision to extend the age range of the participants - I suspect that it is unlikely that the issues that preoccupy a 16 year old will have much in common with those of a 30 year old. I would encourage Melanie to either, (i) stick with an adolescent population or, if necessary, (ii) increase the upper age limit to perhaps 21, to fall within the category of 'young adult'. On that note, the information leaflet talks about 'young people' and I wouldn't class a 30 year old within that bracket.
- The revised project seems to involve analyses using the measures that are not outlined in the research questions section. It would benefit from being clearer about the research question(s) that the project seeks to address.
- The researcher(s) should consider the issue of the internet forum being used by CF patients who do not actually post to the forum. These individuals may benefit from the forum, despite only reading rather than contributing. Arguably individuals with depression or low self-esteem might be more likely to engage in such passive forum use. This is probably just something to consider in the thesis discussion.
- The project may wish to take step(s) to ensure that the person who gives consent is actually the person connected with a given username (e.g. by asking them to post something) prior to using that username's data.

APPENDIX C: CF TRUST AGREEMENT EMAILS

John Devlin

Sent: 01 December 2010 10:55
To: Platten Melanie (NHS DUMFRIES & GALLOWAY)

Hi Melanie,

That sounds fine to me, and as before, we would probably make the initial post from us, to show that it is all legit.

Look forward to hearing from you in the new year with further details.

Best
John

From: Platten Melanie (NHS DUMFRIES & GALLOWAY)
Sent: 30 November 2010 09:57
To: John Devlin
Subject: Research

Hi there John,
I emailed a few weeks ago about changes to my thesis planned project. As I mentioned, the research team felt that my original idea of doing a straight qualitative analysis of the forum posts wasn't strongly enough linked to clinical aspects of my course. After some consideration of alternative ideas which did not use online info, we have had a rethink about the project. I was hoping that it may be possible to still use the CF Trust Forum?

In order to increase the links to clinical practice and to overcome some of the concerns regarding users giving their informed consent for posts to be used in research, we have come up with the idea of asking users to fill in a questionnaire online (via survey monkey) within which would be an information page, consent form and a couple of brief questionnaires. I should have a full research protocol written up in the next week or two which I can send you for more info. I just wanted to check with you to make sure that using the forum would still be a possibility? The vague time line would be to go back to the uni ethics committee in December, finalise project details in January and hopefully collect data from February - April 2011.

Sorry that things have changed around a bit! I hope that you can help and look forward to hearing from you in the near future.
Best wishes,

Melanie Platten
Clinical Psychologist in Training
PPALS, Royal Hospital for Sick Children, 3 Rillbank Terrace, Edinburgh

John Devlin

Sent: 07 February 2011 10:45

To: Platten Melanie (NHS DUMFRIES & GALLOWAY)

Hi Mel,

The page is now up and I will put the post on the forum today.
The page is here: http://www.cftrust.org.uk/aboutcf/livingwithcf/supres
Can you remind me, did you want the teen and adult forums?

John

From: Platten Melanie (NHS DUMFRIES & GALLOWAY)
Sent: 03 February 2011 12:42
To: John Devlin
Subject: Approval letter and updated information page

Hi there John,

I've finally received an official letter from my ethics tutor - please see attached.
I've also attached an updated version of the protocol and information page. We decided to take off the upper age limit for the study to hopefully encourage as many people to take part as possible, so it's now for people aged 16 and up. The information page attached (dated 3rd Feb) is the most recent one and superseeds others. Sorry to keep changing things but I promise this is the final version!

Also, I've added in the website of the survey onto the information page so that people can get started and fill in the survey. For your info, the web address for the survey is:
https://www.surveymonkey.com/s/CF_onlinesocialsupport

When do you think it would be possible to get the information page up on the site and the initial posting? For me, it would be ideal if it could be up by the beginning of next week if at all possible.

I hope to hear from you soon.
Many thanks,

Mel

Melanie Platten
Clinical Psychologist in Training
PPALS, Royal Hospital for Sick Children,
3 Rillbank Terrace, Edinburgh.

APPENDIX D: CLINICAL PSYCHOLOGY REVIEW AUTHOR GUIDELINES

ISSN: 0272-7358
Imprint: ELSEVIER

Article structure
Manuscripts should be prepared according to the guidelines set forth in the Publication Manual of the American Psychological Association (6th ed., 2009).

Manuscripts should ordinarily not exceed 50 pages. Exceptions may be made with prior approval of the Editor in Chief for manuscripts including extensive tabular or graphic material, or appendices.

Appendices
If there is more than one appendix, they should be identified as A, B, etc. Formulae and equations in appendices should be given separate numbering: Eq. (A.1), Eq. (A.2), etc.; in a subsequent appendix, Eq. (B.1) and so on. Similarly for tables and figures: Table A.1; Fig. A.1, etc.

Essential title page information

Title. Concise and informative. Titles are often used in information-retrieval systems. Avoid abbreviations and formulae where possible. **Note: The title page should be the first page of the manuscript document indicating the author's names and affiliations and the corresponding author's complete contact information.**

Author names and affiliations. Where the family name may be ambiguous (e.g., a double name), please indicate this clearly. Present the authors' affiliation addresses (where the actual work was done) below the names. Indicate all affiliations with a lower-case superscript letter immediately after the author's name and in front of the appropriate address. Provide the full postal address of each affiliation, including the country name, and, if available, the e-mail address of each author within the cover letter.

Corresponding author. Clearly indicate who is willing to handle correspondence at all stages of refereeing and publication, also post-publication. **Ensure that telephone and fax numbers (with country and area code) are provided in addition to the e-mail address and the complete postal address.**

Present/permanent address. If an author has moved since the work described in the article was done, or was visiting at the time, a "Present address"' (or "Permanent address") may be indicated as a footnote to that author's name. The address at which the author actually did the work must be retained as the main, affiliation address. Superscript Arabic numerals are used for such footnotes.

Abstract
A concise and factual abstract is required (not exceeding 200 words). This should be typed on a separate page following the title page. The abstract should state briefly the purpose of the research, the principal results and major conclusions. An abstract is often presented separate from the article, so it must be able to stand alone. References should therefore be avoided, but if essential, they must be cited in full, without reference to the reference list.

Highlights
Highlights are mandatory for this journal. They consist of a short collection of bullet points that convey the core findings of the article and should be submitted in a separate file in the online submission system. Please use 'Highlights' in the file name and include 3 to 5 bullet points (maximum 85 characters including spaces, or, maximum 20 words per bullet point). See http://www.elsevier.com/highlights for examples.

Keywords
Immediately after the abstract, provide a maximum of 6 keywords, using American spelling and avoiding general and plural terms and multiple concepts (avoid, for example, "and", "of"). Be sparing with abbreviations: only abbreviations firmly established in the field may be eligible. These keywords will be used for indexing purposes.

Abbreviations
Define abbreviations that are not standard in this field in a footnote to be placed on the first page of the article. Such abbreviations that are unavoidable in the abstract must be defined at their first mention there, as well as in the footnote. Ensure consistency of abbreviations throughout the article.

Acknowledgements
Collate acknowledgements in a separate section at the end of the article before the references and do not, therefore, include them on the title page, as a footnote to the title or otherwise. List here those individuals who provided help during the research (e.g., providing language help, writing assistance or proof reading the article, etc.).

Footnotes
Footnotes should be used sparingly. Number them consecutively throughout the article, using superscript Arabic numbers. Many wordprocessors build footnotes into the text, and this feature may be used. Should this not be the case, indicate the position of footnotes in the text and present the footnotes themselves separately at the end of the article. Do not include footnotes in the Reference list.
Table footnotes
Indicate each footnote in a table with a superscript lowercase letter.

Electronic artwork
General points
• Make sure you use uniform lettering and sizing of your original artwork.
• Save text in illustrations as "graphics" or enclose the font.
• Only use the following fonts in your illustrations: Arial, Courier, Times, Symbol.
• Number the illustrations according to their sequence in the text.
• Use a logical naming convention for your artwork files.
• Provide captions to illustrations separately.
• Produce images near to the desired size of the printed version.
• Submit each figure as a separate file.

A detailed guide on electronic artwork is available on our website:
http://www.elsevier.com/artworkinstructions

Color artwork
Please make sure that artwork files are in an acceptable format (TIFF, EPS or MS Office files) and with the correct resolution. If, together with your accepted article, you submit usable color figures then Elsevier will ensure, at no additional charge, that these figures will appear in color on the Web (e.g., ScienceDirect and other sites) regardless of whether or not these illustrations are reproduced in color in the printed version. **For color reproduction in print, you will receive information regarding the costs from Elsevier after receipt of your accepted article.** Please indicate your preference for color in print or on the Web only. For further information on the preparation of electronic artwork, please see http://www.elsevier.com/artworkinstructions.
Please note: Because of technical complications which can arise by converting color figures to "gray scale" (for the printed version should you not opt for color in print) please submit in addition usable black and white versions of all the color illustrations.

Figure captions
Ensure that each illustration has a caption. Supply captions separately, not attached to the figure. A caption should comprise a brief title (**not** on the figure itself) and a description of the illustration. Keep text in the illustrations themselves to a minimum but explain all symbols and abbreviations used.

Tables
Number tables consecutively in accordance with their appearance in the text. Place footnotes to tables below the table body and indicate them with superscript lowercase letters. Avoid vertical rules. Be sparing in the use of tables and ensure that the data presented in tables do not duplicate results described elsewhere in the article.

References
Citations in the text should follow the referencing style used by the American Psychological Association. You are referred to the Publication Manual of the American Psychological Association, Sixth Edition, ISBN 1-4338-0559-6, copies of which may be ordered from http://books.apa.org/books.cfm?id=4200067 or APA Order Dept., P.O.B. 2710, Hyattsville, MD 20784, USA or APA, 3 Henrietta Street, London, WC3E 8LU, UK. Details concerning this referencing style can also be found at http://humanities.byu.edu/linguistics/Henrichsen/APA/APA01.html

Citation in text
Please ensure that every reference cited in the text is also present in the reference list (and vice versa). Any references cited in the abstract must be given in full. Unpublished results and personal communications are not recommended in the reference list, but may be mentioned in the text. If these references are included in the reference list they should follow the standard reference style of the journal and should include a substitution of the publication date with either "Unpublished results" or "Personal communication" Citation of a reference as "in press" implies that the item has been accepted for publication.

Web references
As a minimum, the full URL should be given and the date when the reference was last accessed. Any further information, if known (DOI, author names, dates, reference to a source publication, etc.), should also be given. Web references can be listed separately (e.g., after the reference list) under a different heading if desired, or can be included in the reference list.

References in a special issue
Please ensure that the words 'this issue' are added to any references in the list (and any citations in the text) to other articles in the same Special Issue.

Reference management software
This journal has standard templates available in key reference management packages EndNote (http://www.endnote.com/support/enstyles.asp) and Reference Manager (http://refman.com/support/rmstyles.asp). Using plug-ins to wordprocessing packages, authors only need to select the appropriate journal template when preparing their article and the list of references and citations to these will be formatted according to the journal style which is described below.

Reference style

References should be arranged first alphabetically and then further sorted chronologically if necessary. More than one reference from the same author(s) in the same year must be identified by the letters "a", "b", "c", etc., placed after the year of publication. **References should be formatted with a hanging indent (i.e., the first line of each reference is flush left while the subsequent lines are indented).**

Examples: Reference to a journal publication: Van der Geer, J., Hanraads, J. A. J., & Lupton R. A. (2000). The art of writing a scientific article. *Journal of Scientific Communications,* 163, 51-59.

Reference to a book: Strunk, W., Jr., &White, E. B. (1979). *The elements of style.* (3rd ed.). New York: Macmillan, (Chapter 4).

Reference to a chapter in an edited book: Mettam, G. R., & Adams, L. B. (1994). How to prepare an electronic version of your article. In B.S. Jones, & R. Z. Smith (Eds.), *Introduction to the electronic age* (pp. 281-304). New York: E-Publishing Inc.

Supplementary data
Elsevier accepts electronic supplementary material to support and enhance your scientific research. Supplementary files offer the author additional possibilities to publish supporting applications, high-resolution images, background datasets, sound clips and more. Supplementary files supplied will be published online alongside the electronic version of your article in Elsevier Web products, including ScienceDirect: http://www.sciencedirect.com. In order to ensure that your submitted material is directly usable, please provide the data in one of our recommended file formats. Authors should submit the material in electronic format together with the article and supply a concise and descriptive caption for each file. For more detailed instructions please visit our artwork instruction pages at http://www.elsevier.com/artworkinstructions.

Submission checklist
The following list will be useful during the final checking of an article prior to sending it to the journal for review. Please consult this Guide for Authors for further details of any item.
Ensure that the following items are present:
One Author designated as corresponding Author:
• E-mail address
• Full postal address
• Telephone and fax numbers
All necessary files have been uploaded
• Keywords
• All figure captions
• All tables (including title, description, footnotes)
Further considerations
• Manuscript has been "spellchecked" and "grammar-checked"
• References are in the correct format for this journal
• All references mentioned in the Reference list are cited in the text, and vice versa
• Permission has been obtained for use of copyrighted material from other sources (including the Web)
• Color figures are clearly marked as being intended for color reproduction on the Web (free of charge) and in print or to be reproduced in color on the Web (free of charge) and in black-and-white in print
• If only color on the Web is required, black and white versions of the figures are also supplied for printing purposes
For any further information please visit our customer support site at http://support.elsevier.com.

APPENDIX E: COPY OF ONLINE SURVEY FOR STUDY 1

APPENDIX F: EMAIL PERMISSIONS FOR STANDARDISED MEASURES

Matt Brown
Sent: 10 December 2010 09:29
To: Platten Melanie (NHS DUMFRIES & GALLOWAY)

Good Morning

I don't believe that there is an issue with the transferring of the CFQ-R.

This should be with you early next week.

I will have to just double check with Mandy Bryon at GOSH before I send this out to you.

Kind Regards

Matthew Brown
Product Manager

Forest Laboratories UK Ltd
Riverbridge House
Anchor Boulevard
Crossways Business Park
Dartford, Kent
DA2 6SL

From: Platten Melanie (NHS DUMFRIES & GALLOWAY)
Sent: 09 December 2010 17:57
To: Matt Brown
Subject: RE: CFQ-R permissions

Hi there Matthew,

Thanks for getting back to me and for sending through the information. My address is at the bottom of this email if you could send things there.

Can I take it that there is no copyright problem with transferring the CFQ-R onto my surveymonkey online survey for my doctoral thesis?

Thanks,

Melanie Platten
Clinical Psychologist in Training

PPALS, Royal Hospital for Sick Children,
3 Rillbank Terrace, Edinburgh, EH9 1LL.

From: Matt Brown
Sent: 09 December 2010 11:00
To: Platten Melanie (NHS DUMFRIES & GALLOWAY)
Subject: CFQ-R permissions

Good Morning,

Following on from our discussion, I shall be sending a CFQ pack to you. In addition, there are some attachments to this email which help with the CFQ.

First is the CFQ presentation given by Mandy Bryon from GOSH in April 2009. This helps explain the CFQ & in addition the overview PDF.

The pack uses the CFQ (USA version) of the disc. This has a glitch in it, which isn't anything we can reverse, (as it wasn't a Forest disc to start with).

The word document should help explain this.

Can you confirm the full address you wish the CFQ to go to please?

Kind Regards

Matthew Brown
Product Manager

Forest Laboratories UK Ltd
Riverbridge House
Anchor Boulevard
Crossways Business Park
Dartford, Kent
DA2 6SL

Rebecca Hoskin

Sent: 20 December 2010 09:48

To: Platten Melanie (NHS DUMFRIES & GALLOWAY)

Cc: Leanne Robotham

Dear Melanie,

Please see response from our Managing Director below,

Could you please permission this request on my behalf and explain that Richard Evans is off ill.

Please explain that permission is granted only for the purpose of the Doctoral research project

Thank you

John

If we can be of any further assistance please do not hesitate in contacting me.

Best Wishes
Rebecca Hoskin – Customer Support
tel: 01788 546019
fax: 01788 331407

Please note the CORE administration office will be closed from 24th December and re-open Tuesday 04th January 2011.

All the team at CORE IMS wish you a very happy holiday season and all the best for 2011

-----Original Message-----
From: Platten Melanie (NHS DUMFRIES & GALLOWAY)
Sent: 14 December 2010 11:27
To: Admin
Subject: RE: CORE System Forms

Hi there,

I recently tried get in touch with you (via riche@btclick.com) to ask permission to use the CORE-34 items electronically for my Doctoral research project. My thesis project will be carried out online and participants recruited via a website and then completing a survey online. I wondered whether it would be possible to gain permission to transfer the items of the CORE onto my online survey for use within this context? I would, of course, reference the questionnaire appropriately and maintain the order, wording and ratings of each items as on the forms. The survey will make use of survey monkey for ease of creation and collection of data. It is hoped that data collection using the survey will be open for participants between February to April 2011. I am about submit my project to an ethics committee and would very

much like to use the CORE if at all possible. I do not have a budget for my project so hope that there would be no large fee for doing this if permission was granted.

Thanks and I look forward to hearing from you in the near future.

Yours faithfully,

Melanie Platten
Clinical Psychologist in Training

PPALS, Royal Hospital for Sick Children,
3 Rillbank Terrace, Edinburgh.

_____

From: admin@coreims.co.uk
Sent: 07 December 2010 15:41
To: Platten Melanie (NHS DUMFRIES & GALLOWAY)
Subject: CORE System Forms

Thank you for your enquiry to CORE Information Management Systems (CORE IMS) requesting CORE System forms. The forms you requested are attached.

Please note that these are for your own use in connection with your therapy practice, service or research, as outlined in the Conditions of Use statement. Please do not resend this email or the forms electronically. If you know others who may wish to access the forms please direct them to the Resources section of our website www.coreims.co.uk.

CORE IMS is the sole organisation licensed by the CORE System Trust to provide support for CORE System users. This includes training in implementing the CORE System and using CORE System data in practitioner and service development. CORE IMS also provides CORE PC software to support the development of excellence in practice and service delivery – visit our website www.coreims.co.uk for further details

APPENDIX G: INITIAL INDEX FRAMEWORK FOR QUALITATIVE ANALYSIS

Version 1
Date: 19th May 2011

Forum Post - Qualitative Indexing Framework

1. CF treatment
- 1.1. Nebulisers, antibiotics and doseage
- 1.2. Length of time to take treatments
- 1.3. Fitting in treatment into everyday life
- 1.4. Pro's and con's of different treatment methods
- 1.5. Highlighting the important of treatments
- 1.6. Side-effects
- 1.7. Lung transplant
- 1.8. New developments in treatments
- 1.9. Inpatient stays
- 1.10. CF Unit facilities
- 1.11. Exercise tests
- 1.12. Segregation
- 1.13. Other

2. Medical procedures
- 2.1. Blood tests
- 2.2. IV antibiotics
- 2.3. Ports
- 2.4. Pain
- 2.5. Fear or panic
- 2.6. Ways of coping and tips for comfort
- 2.7. Inexperience medical staff
- 2.8. Other

3. Health topics
- 3.1. Improvements in health
- 3.2. Health advice seeking
- 3.3. Information about aspects of non-CF health e.g. migraines, dental
- 3.4. Respiratory Infections
- 3.5. Effects of pregnancy
- 3.6. Menstruation
- 3.7. Speaking to health team
- 3.8. Other

4. Daily living issues and occupation
- 4.1. Education inc. school and university attendance or exams
- 4.2. Work and employment
- 4.3. Interviews – disclosing diagnosis
- 4.4. Practical tips about comfortable clothing
- 4.5. Mobility / cars
- 4.6. DLA and benefits
- 4.7. Smokers in public
- 4.8. Fitness and running
- 4.9. Non-CF issues e.g. heating at home
- 4.10. Past-times and hobbies
- 4.11. Other

5. Psychosocial and inter-personal relationships
- 5.1. Self-esteem
- 5.2. Adjusting to changes in health / treatment
- 5.3. Self-help services and groups
- 5.4. Friends not understanding
- 5.5. Feeling different or "special"
- 5.6. Hiding symptoms from others e.g. suppressing cough
- 5.7. Other

6. Future
- 6.1. Fear of what the future holds
- 6.2. Hope
- 6.3. Pregnancy, fertility and genetics
- 6.4. Parenting with CF
- 6.5. Morality of having children
- 6.6. Other

7. Nature of post
- 7.1. Encouragement
- 7.2. Reassurance
- 7.3. Empathy
- 7.4. Sharing experiences
- 7.5. Giving information or advice
- 7.6. Providing opinion or alternative viewpoint
- 7.7. Seeking support
- 7.8. Giving condolences after a death
- 7.9. Well-wishing
- 7.10. Thanks
- 7.11. Use of humour
- 7.12. Clarifying tone / intention
- 7.13. Diffuse potential arguments
- 7.14. Other

8. Other key issues (not covered above)
- 8.1. CF related
- 8.2. Non-CF related

APPENDIX H: FINAL INDEX FRAMEWORK FOR QUALITATIVE ANALYSIS

Version 4
Date: 29th May 2011

Forum Post - Qualitative Indexing Framework

1. CF treatment
- 1.1. Nebulisers, antibiotics and doseage
- 1.2. Physiotherapy
- 1.3. Exercise tests
- 1.4. Lung transplant
- 1.5. Fitting in treatment into everyday life
- 1.6. Pro's and con's of different treatment methods
- 1.7. Side-effects
- 1.8. Highlighting the important of treatments
- 1.9. CF Unit facilities
- 1.10. Segregation

2. Medical procedures
- 2.1. Blood tests
- 2.2. IV antibiotics
- 2.3. Ports
- 2.4. Bronchoscopy
- 2.5. Diagnosis
- 2.6. Health staff skills

3. Health topics
- 3.1. Improvements or exacerbations in health
- 3.2. Health advice seeking
- 3.3. CF complications
- 3.4. Physical development
- 3.5. Diet, enzymes and digestion
- 3.6. Speaking to health team
- 3.7. Information about aspects of non-CF health e.g. migraines, dental, flu jab
- 3.8. Home adjustments (e.g. showerheads, air purifier)

4. Daily living issues and occupation
- 4.1. Education inc. school and university attendance or exams
- 4.2. Employment and interviews
- 4.3. DLA, mobility and benefits
- 4.4. Smokers
- 4.5. Fitness and running
- 4.6. Non-CF issues e.g. heating at home
- 4.7. Past-times and hobbies
- 4.8. Fundraising
- 4.9. Travel insurance and healthcare abroad

5. Psychosocial processes and issues
- 5.1. Adjustment and transitions
- 5.2. Friends not understanding
- 5.3. Feeling different or "special"
- 5.4. Hiding symptoms from others e.g. suppressing cough
- 5.5. Emotions inc. low mood, anxiety, fear
- 5.6. Anger at illness and other's waste of health
- 5.7. Coping
- 5.8. Body image and weight
- 5.9. "Invisible" illness
- 5.10. Psychologists, self-help and psychology services

6. Future
- 6.1. Fear of what the future holds
- 6.2. Hope
- 6.3. Pregnancy, fertility and genetics
- 6.4. Parenting
- 6.5. Morality of having children
- 6.6. Politics and changes in health service

7. Nature of post
- 7.1. Giving condolences after a death
- 7.2. Well-wishing
- 7.3. Use of humour
- 7.4. Diffuse potential arguments

All posts have general tone of:
- 7.5. Encouragement
- 7.6. Reassurance
- 7.7. Empathy
- 7.8. Sharing experiences
- 7.9. Giving information or advice
- 7.10. Providing opinion or alternative viewpoint
- 7.11. Seeking support
- 7.12. Thanks
- 7.13. Clarifying tone / intention

APPENDIX I: EXAMPLE OF A FRAMEWORK ANALYSIS DATA MATRIX

1. CF TREATMENT

Username, Gender, Age, Ethnicity	a) Nebulisers, antibiotics and dosage	b) IV anitbiotics	c) Blood Tests
P1; Female, 25yo	Providing info on Tobra and influential factors on dosage (weight, kidney function). Also giving info on nebs (order of meds, "prettied up")	Coincides with periods, body has a sixth sense. Makes IVs more difficult to cope with. Feel the benefit after 7 days, next week clears up. Sometimes not as effective though	Taking Tobi bloods from port instead of regular veins - same results. Empathising with frustration when bloods get lost etc. Painful re-attempts, some staff better than others. Hypnotherapy helped when younger - needed restraining, now suck thumb for comfort when they jab me.
P2; Female, 29yo			
P3; Female, 25yo		Seeking advice bronchoscopy - whether more IVs needed afterwards. Providing experience of feeling better after few days of Ivs, trusting instincts on need for treatment.	
P4; Female, 24yo			
P5; Male, 32yo		Frequency of IVs decreased since stopping work, cost less to society. Impact of IVs can vary, days or weeks to feel it. Sometimes feel rubbish doing Ivs. Wee smelling!	
P6; Male, 31yo	Using salbutamol and oral abs to combat cold. Sharing to see if others have input on possible problems		
P7; Female, 28yo	Seeking help with and discussing dosage of Tobra. Sharing experience of saline to hydrate lungs. Unable to share if funny taste due to cold!	Port makes it easier to get access for IVs compared to Venflon, put it off for 10 years. Veins burst = lumps, anesthetist better at putting in	
P8; Female, 26yo	Explaining hypertonic saline, concentrations and mechanisms of action. Asking for different abs at clinic		Giving blood not a big deal, it's a pain especially when they miss, hit nerves, lose vials but it's necessary. Ports can be easier if hate needles.
P9; Female, 27yo	Requirement for blood tests when taking Tobra		Tobra dose check done via finger prick, few pricks needed to get enough blood.
P10; Male, 38yo		Developed reactions to most IVs, cumulative effect over the years.	

APPENDIX J: JOURNAL OF CYSTIC FIBROSIS AUTHOR GUIDELINES

The Official Journal of the European Cystic Fibrosis Society

Journal of Cystic Fibrosis publishes original scientific articles, editorials, case reports, short communications and other information relevant to cystic fibrosis and is published four times a year. Accepted papers become the copyright of the Journal and are accepted on the understanding that they have not been published, and are not being considered for publication elsewhere and are subject to editorial revision.

Online Submission of Manuscripts to the *Journal of Cystic Fibrosis:*
Submission and peer review of all papers is now conducted entirely online, increasing efficiency for editors, authors, and reviewers, and enhancing publication speed. Submit online at: http://ees.elsevier.com/jcf/
Authors are guided stepwise through the entire process, and are kept abreast of the progress of their paper at each stage. The system creates PDF version of the submitted manuscript for peer review, revision and proofing. All correspondence, including the Editor's decision and request for revisions, is conducted by e-mail.
Authors requesting further information on online submission are strongly encouraged to view the system, including a tutorial, at http://ees.elsevier.com/jcf. A comprehensive Author Support service is also available to answer additional enquiries at authorsupport@elsevier.com.

Cover letter
Corresponding authors must provide a cover letter which includes statements answering the following questions:

- Has the work been seen and approved by all co-authors?
- How is the work clinically relevant, and how does it add to existing research?
- Have papers closely related to the submitted manuscript been published or submitted for publication elsewhere? If so please provide details.

Failure to provide a cover letter addressing each of the questions above will result in the paper being returned to the author. The cover letter must be uploaded as a separate submission item.

Title
Your title page, numbered as 1, should give the title in capital letters (not exceeding 100 letters), a running title (not exceeding 50 letters) and the authors' names (as they are to appear), affiliations and complete addresses, including postal (zip) codes. The author and address to whom correspondence should be sent must be clearly indicated. Please supply telephone, fax and e-mail numbers for the corresponding author. Avoid use of extraneous words such as "study", "investigation", etc. If data from the manuscript have been presented at a meeting, list the full name, date and location of the meeting and reference any previously abstracts in the bibliography.

Abstracts
An abstract of your manuscript, summarizing the content, should be provided. A maximum of 150 words, should be written in a structured manner (for original articles only) since this will be the only part of the article studied by some readers. In original articles, the Abstract should consist of 4 paragraphs, labelled Background, Methods, Results, and Conclusions. They should briefly describe the problem being addressed in the study, how the study was performed and which measurements were carried out, the most relevant results, and what the authors conclude from the results.

Reference Format
Manuscripts should use the 'Vancouver' style for references, which should be numbered consecutively (in parentheses) in the order in which they are first cited in the text and listed at the end of the paper. For journal references, all authors should be included when there are six or less (first three only when seven or more), followed by the title of article, name of journal abbreviated according to British Standard 4148: 1975 (or left in full), year, volume, and first and last pages.
For example:
1. Tockman MS, Anthonisen MD, Wright EC et al. Airways obstruction and the risk of lung cancer. Ann Intern Med 1987; 106:512-518.
For book references, the author(s) should be followed by the chapter title (if appropriate), editor(s) (if applicable), book title, place of publication, publisher, year and page numbers.
For example:
2. Colby VT, Carrington CB. Infiltrative lung disease. In: Thurlbeck WM, ed. Pathology of the Lung. New York: Thieme Medical Publishers, 1988.

Figures
Figures of good quality should be submitted online as a separate file. For detailed instructions on the preparation of electronic artwork, consult: http://www.elsevier.com/authors. Permission to reproduce illustrations should always be obtained before submission and details included with the captions.

Tables

Tables should be typed with double spacing each on a separate file, numbered consecutively with Arabic numerals, and contain only horizontal lines. Provide a short descriptive heading above each table with footnotes and/or explanations underneath.

Keywords
A list of three to six keywords should be supplied: full instructions are provided when submitting the article online.

Types of submission

Short Communication
1,200 words plus no more than 3 figures or tables in total 20 references
Original articles
Original research papers should contain no more than 3,000 words plus no more than 5 figures or tables in total and 30 references. The abstract should consist of 4 paragraphs, labelled Background, Methods, Results, and Conclusions.
Review articles
Review papers should be authoritative, well referenced reviews of a relevant subject and should not contain more than 5,000 words, and 30 references with no more than six figures or tables.

Units & Abbreviations
These should be given in SI units with the traditional equivalent in parentheses where appropriate. All other abbreviations or acronyms should be defined on first appearance in the text. Use a capital letter for proprietary names of substances and materials. At first mention of a chemical substance, use the correct chemical designation as well as the generic name.

Authorship
All authors should have made substantial contributions to all the following: (1) the conception and design of the study, or acquisition of the data, or analysis and interpretation of data, (2) drafting or revising it critically for important intellectual content, (3) final approval of the version to be submitted.

Acknowledgements
All contributors who do not meet the criteria for authorship as defined above should be listed in an acknowledgements section. Examples of those who might be acknowledged include a person who provided purely technical help, writing assistance, or a department chair who provided only general support. Authors should disclose whether they had any writing assistance and identify the entity that paid for this assistance.

Conflict of interest
The potential for conflict of interest exists when an author (or the author's institution), reviewer or editor has financial or personal relationships that may inappropriately influence his or her actions. Editors and reviewers for the journal are responsible for disclosing to the Chief Editor any personal or financial relationship that may bias their work during the peer review process.
Authors are specifically asked to reflect on financial conflicts of interest (such as employment, consultancy, stock ownership, honoraria and paid expert testimony) as well as other forms of conflict of interest, including personal, academic and intellectual issues.
At the end of the text, under a subheading "Conflict of interest statement" all authors must disclose any financial and personal relationships with other people or organisations that could inappropriately influence (bias) their work.
Examples of potential conflicts of interest include employment, consultancies, stock ownership, honoraria, paid expert testimony, patent applications/registrations, and grants or other funding.

Role of the funding source
All sources of funding should be declared as an acknowledgement at the end of the text. Authors should declare the role of study sponsors, if any, in the study design, in the collection, analysis and interpretation of data; in the writing of the manuscript; and in the decision to submit the manuscript for publication. If the study sponsors had no such involvement, the authors should so state.

Randomised controlled trials
All randomised controlled trials submitted for publication in the Journal of Cystic Fibrosis should include a completed Consolidated Standards of Reporting Trials (CONSORT) flow chart. Please refer to the CONSORT statement website at http://www.consort-statement.org for more information. Journal of Cystic Fibrosis has adopted the proposal from the International Committee of Medical Journal Editors (ICMJE) which require, as a condition of consideration for publication of clinical trials, registration in a public trials registry. Trials must register at or before the onset of patient enrolment. The clinical trial registration number should be included at the end of the abstract of the article. For this purpose, a clinical trial is defined as any research study that prospectively assigns human participants or groups of humans to one or more health-related interventions to evaluate the effects of health outcomes. Health-related interventions include any intervention used to modify a biomedical or health-related outcome (for example drugs, surgical procedures, devices, behavioural treatments, dietary interventions, and process-of-care changes). Health outcomes include any biomedical or health-related measures obtained in patients or participants, including pharmacokinetic measures and adverse events. Purely observational studies (those in which the assignment of the medical intervention is not at the discretion of the investigator) will not require registration. Further information can be

found at http://www.icmje.org

Disclosure of Clinical Trial Results
In line with the position of the International Committee of Medical Journal Editors , the journal will not consider results posted in the same clinical trials registry in which primary registration resides to be prior publication if the results posted are presented in the form of a brief structured (less than 500 words) abstract or table. However, divulging results in other circumstances (eg, investors' meetings) is discouraged and may jeopardise consideration of the manuscript. Authors should fully disclose all posting in registries of results of the same or closely related work.

Ethics
Work on human beings that is submitted to Respiratory Medicine should comply with the principles laid down in the Declaration of Helsinki; Recommendations guiding physicians in biomedical research involving human subjects. Adopted by the 18th World Medical Assembly, Helsinki, Finland, June 1964, amended by the 29th World Medical Assembly, Tokyo, Japan, October 1975, the 35th World Medical Assembly, Venice, Italy, October 1983, and the 41st World Medical Assembly, Hong Kong, September 1989. The manuscript should contain a statement that the work has been approved by the appropriate ethical committees related to the institution(s) in which it was performed and that subjects gave informed consent to the work. Studies involving experiments with animals must state that their care was in accordance with institution guidelines. Patients' and volunteers' names, initials, and hospital numbers should not be used.

Copyright
Accepted papers become the copyright of the Journal and are accepted on the understanding that they have not been published, are not being considered for publication elsewhere and are subject to editorial revision. If papers closely related to the submitted manuscript have been published or submitted for publication elsewhere, the author must state this in their cover letter Upon acceptance of an article, authors will be asked to sign a "Journal Publishing Agreement" (for more information on this and copyright see http://www.elsevier.com/authors). Acceptance of the agreement will ensure the widest possible dissemination of information. An e-mail (or letter) will be sent to the corresponding author confirming receipt of the manuscript together with a 'Journal Publishing Agreement' form.
If excerpts from other copyrighted works are included, the author(s) must obtain written permission from the copyright owners and credit the source(s) in the article. Elsevier has preprinted forms for use by authors in these cases : contact e-mail healthpermissions@elsevier.com. Requests may also be completed online via the Elsevier homepage (http://www.elsevier.com/locate/permissions).

Permissions Information
If illustrations are borrowed from published sources, written permission must be obtained from both publisher and author, and a credit line giving the source added to the legend. If text material totalling 250 to 300 words, or any tables, are borrowed verbatim from published sources, written permission is required from both publisher and author. With shorter quotations, it is sufficient to add a bibliographic credit. Permission letters for reproduced text or illustration must accompany the manuscript. If you have been unable to obtain permission, please point this out.

Proofs
Proofs will be sent to the authors to be carefully checked for printer's errors. Changes or additions to the edited manuscript cannot be allowed at this stage. Corrected proofs should be returned to the publisher within 2 days of receipt.

Reprints/Author Benefits
The *Journal of Cystic Fibrosis* has no page charges. Reprints. Twenty Five free reprints will be supplied per article. The publisher will send authors a form enabling further reprints to be ordered at prices listed on the form.

Accepted Articles
For the facility to track accepted articles and set email alerts to inform you of when an article's status has changed visit http://authors.elsevier.com/TrackPaper.html. There are also details on artwork guidelines, copyright information, frequently asked questions and more. Contact details for questions arising after acceptance of an article, especially those related to proofs, are provided after registration of an article for publication.

Funding body agreements and policies
Elsevier has established agreements and developed policies to allow authors who publish in Elsevier journals to comply with potential manuscript archiving requirements as specified as conditions of their grant awards. To learn more about existing agreements and policies please visit http://www.elsevier.com/fundingbodies.